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A rare case of co-secreting adrenocortical oncocytoma

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A B S T R A C T

Here we present a rare case of a large co-secreting adrenocortical oncocytoma undergoing laparoscopic retroperitoneal-to-open removal. Our case was referred for surgical opinion as a result of a left suprarenal mass being identified as part of a work-up for new onset hirsutism and post-menopausal bleeding. We provide example of our surgical approach as well as discussion on this unique adrenal tumour variant.

1. Introduction

Adrenal oncocytoma is an uncommon pathology and is usually identified incidentally on cross-sectional imaging. Within this report we describe our case of a large functional adrenal oncocytoma, a rare tumour, which was identified on investigation of hirsutism and post-menopausal bleeding. We outline our initial work-up of endocrine investigation and discuss our surgical approach to the case.

2. Case summary

A 63-year-old female was referred for surgical opinion with new onset hirsutism and post-menopausal bleeding, with CT demonstrating an 8 × 8 × 9.2cm left suprarenal mass (Fig. 1). The patient was recently diagnosed with hypertension and had history of mood disorder with prior inpatient mental health admissions. Examination demonstrated male pattern hair growth to the face, arms, and torso. There were no overt cushingoid features, however she was hypertensive at 150/80 mmHg. The patient had no prior surgical history and was an ex-smoker.

An endocrine panel demonstrated a co-secreting functional tumour, with results as follows: Testosterone 9.3nmol/L (normal range 0–1.5), SHBG 56nmol/L (15–70), free androgen index 19.3% (0.3–5.5), DHEA-S 9.5 μmol/L (1.2–8.4), 17-hydroprogesterone 12.8nmol/L (0.1–1.3), FSH 3.7IU/L (normal range post-menopause 25–130), LH 1.5IU/L (5.0–62), and cortisol 255nmol/L post 1mg dexamethasone suppression test indicating a positive test. Aldosterone could not be accurately assessed as the patient had recently been commenced on spironalactone for hypertension. Pelvic ultrasound and CT chest were unremarkable.

A laparoscopic 4-port retroperitoneal approach was performed with clipping and division of the single adrenal vein and near complete dissection performed prior to planned open completion of the lateral and superior dissection lines to ensure no tumour spillage. Laparoscopic approach was initially conducted in favour of open surgery due to surgeon preference in mobilising the mass away from surrounding structures, which in this case we feel helped distinguish surgical planes whilst minimising blood loss and procedural time. Laparoscopic flank port sites were connected on transition to open surgery. The patient was discharged home on Day 6 with Endocrine review to assist with steroid dosing regimen.

Histopathology revealed an adrenocortical neoplasm of unknown malignant potential according to the Lin-Weiss-Besceglia criteria. The specimen was a well circumscribed, encapsulated tan lesion 97 × 87 × 64mm and weighed 344g (Fig. 2). The cut surface of the specimen was bright yellow, with other fleshy/tan areas and zones of haemorrhage (Fig. 3). Microscopically the specimen consisted of sheets of oncocytic cells with abundant pink to vacuolated cytoplasm, enlarged nuclei and prominent nucleolin (Image 3). There was no convincing capsular invasion, and less than 5mitosis/50HPF with no atypical mitosis identified. Large areas of necrosis and haemorrhage were identified with focal calcification, however there was no vascular or sinusoidal space invasion. Immunochemistry was positive for: MelanA, calretinin, synaptophysin, and inhibin.

Upon review in our post-operative clinic 6-weeks later, the patient had recovered well from surgery and was on a weaning course of hydrocortisone. Repeat investigations revealed a normal endocrine panel. Decision was made following multidisciplinary team discussion for repeat CT imaging at 6-months, followed by 12-monthly imaging for 5-years for oncosurveillance.

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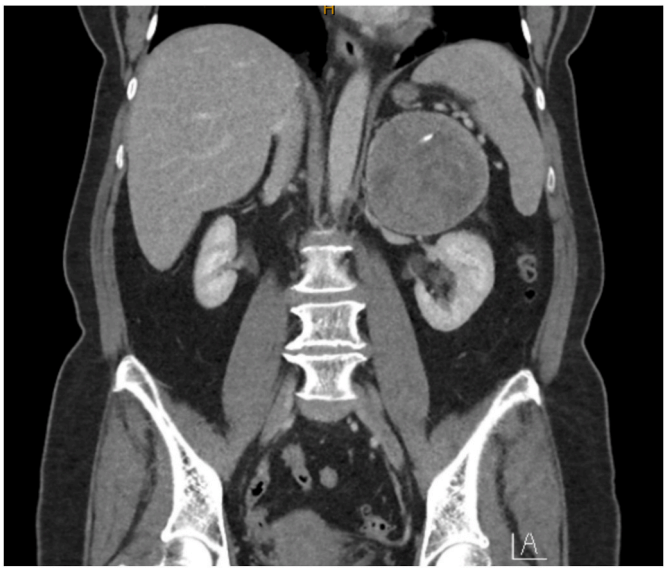

Fig. 1. Coronal CT of an 8 × 8 × 9.2cm left suprarenal mass.

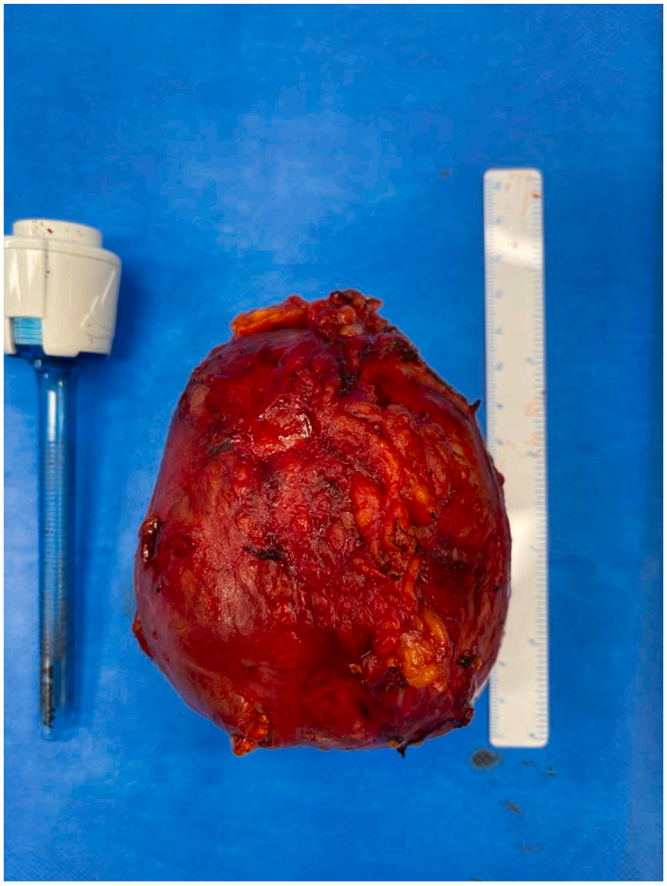

Fig. 2. Post-operative uncut specimen.

3. Discussion

Adrenocortical oncocytoma is a rare pathology that usually presents as an incidental finding on cross sectional imaging and is found in 1.8% of adrenalectomy specimens.[1] This case will add to the literature of functional tumours, of which only 20 have previously been identified, and will assist future management and surveillance decisions in such cases.[2]

First identified in 1986, less than 200 cases of adrenal oncocytoma have been reported in the literature.[3] Studies have shown a 2.5:1 female distribution and 3.5:1 left-sided predominance.[1] Systematic review has identified that only 10–20% of cases are functional tumours, which present with cushingoid and/or virilisation features as a result of lesions usually arising from the adrenal cortex.[1,4] Of published cases 20% were malignant, which portends a poor prognosis of 15–60% 5-year-survival.[4]

The oncocyte was first described in 1950 by Hamperl in relation to the Hurthle-cell thyroid tumour, and describes a pattern of granular, highly eosinophilic cytoplasmic cells resulting from an accumulation of mitochondria as the dominant cell type.[1,3] These tumours most frequently are found within the kidney, parathyroid, pituitary, salivary glands, and the thyroid.[1,2]

Disease classification is based on the Lin-Weiss-Bisceglia scoring system, which provides groupings of disease based on major and minor criteria. Classification is as follows: Malignant (any major criteria), unknown malignant potential (only minor criteria), and benign (no major or minor criteria).[5] Our case was classified into the unknown malignant potential category as a result of fulfilling two minor criteria, in that the tumour weighed >200g and had scattered necrosis.

Operative approach for our case was laparoscopic retroperitoneal, with the patient counselled pre-operatively that there would be a planned open conversion due to tumour size. Whilst recent published guidelines by the European Society for Medical Oncology (ESMO) counsel for open transperitoneal surgery for tumours >8cm in size, systematic review has demonstrated comparative recurrence and disease-free survival rates for open vs. laparoscopic surgery in adrenalectomy, and advised cases should be approached on an individual basis with primary consideration of surgical experience and preference.[4]

Surveillance schedules for adrenocortical neoplasms of unknown potential are absent in the literature due to the rarity of such cases. Prior case reports have identified an 8% recurrence and 3% tumour related mortality rate.[1] As such, following multidisciplinary discussion, our patient will be re-imaged by CT at the 6-month mark, and 12-monthly for the next 5-years providing there is no evidence of recurrence.

4. Conclusion

This rare case adds to the limited published literature of large functioning adrenocortical oncocytoma. The knowledge of variant pathology cases must improve so as to further facilitate evidence-based treatment. Our case identified a tumour of unknown malignant potential, and given the paucity of data to date ongoing surveillance with imaging will be performed.

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The first author, Dr James L. Kovacic, is an Unaccredited Surgical Registrar of Royal North Shore Hospital, Sydney, Australia. Content of this case report manuscript has been reviewed and agreed upon by each of the listed authors.

Written informed consent has been obtained from the patient for completion of this manuscript. This manuscript has not been published nor is it under consideration by another institution.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

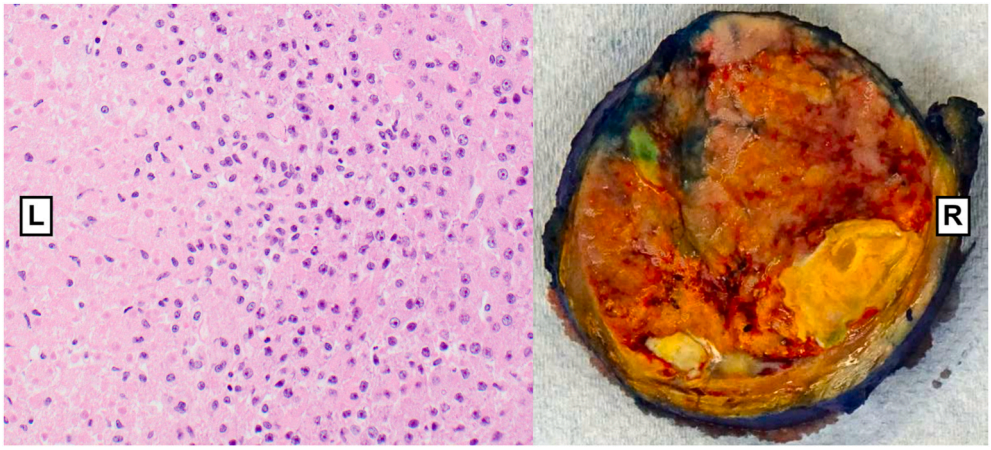

Fig. 3. Left - Microscopic image of specimen. Area of necrosis identified toward left of the slide, with transition to layered sheets of oncocytic cells toward the right. Right - Macroscopic cut image of specimen with bright yellow tissue and other fleshy/tan areas and zones of haemorrhage. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

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