Case report

Adrenocortical oncocytoma: Review of imaging and histopathological implications

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A 31 year-old woman was referred to the hepato-biliary unit after a large right sided abdominal mass was incidentally detected on a CT scan while being investigated for haematuria. Apart from a mild back ache and some early satiety there were no other specific clinical complaints. She was normotensive, fit and well with the normal range. Tumours markers were not raised.

All the blood tests including liver function tests were within the normal range. There was no history suggestive of liver disease and she was a non smoker with a minimal alcohol intake. Beating weight was within the normal range. There were no complications or any sign of adrenal insufficiency.

The CT scan (Fig. 1) revealed a 10 cm × 9 cm × 9 cm mass which was of hypoattenuation consistent with necrosis. The remaining liver parenchyma was normal. A gadolinium-enhanced MRI (Fig. 2) scan was organised to further elucidate the nature of the lesion. This showed features indicative of primary liver tumours such as enhancement in the arterial phase followed by contrast washout in the portal venous phase. However the right adrenal gland could not be identified separately from the lesion. On both imaging modalities a differential diagnosis of primary hepatic tumour or giant adrenal neoplasm was made. Subsequently, plasma and 24-h urinary metanephrines and full adrenal functional profile was measured which was normal.

At laparotomy, the mass was found to be abutting but separate from the liver. Adrenal gland could not be separately identified. The lesion was completely excised. In the post-operative period there were no complications or any sign of adrenal insufficiency. The histology revealed adrenal oncocytoma. She remains well three months later. There is a plan for follow up with interval CT imaging.
given the uncertainty over the long term malignant potential of
these neoplasms.1

3. Histology

Macroscopic examination showed a 10.5 cm × 9.5 cm × 9 cm
rounded, smooth mass weighing 547 grams. The cut surface was
brown and homogenous with a thick fibrous capsule. A compressed
rim of adrenal gland was identified at one edge of the tumour
(Fig. 3).

Microscopic findings showed a well-circumscribed partly
encapsulated tumour composed of nests and trabeculae of large
polygonal cells with abundant eosinophilic granular cytoplasm
(Fig. 4). There was random nuclear atypia with pleomorphic nuclei
containing prominent nucleoli. No mitotic figures were identified.
There was no evidence of capsular or vascular invasion. Residual
adrenal gland was identified adjacent to the tumour and appeared
unremarkable.

The tumour cells were diffusely and strongly positive for Cal-
retinin and Melan A (adrenal cortical markers). There was strong
expression of synaptophysin and patchy but strong chromo-
granin A positivity. Renal and hepatic markers were negative. The
tumour was negative for TTF1 and inhibin. The MIB index was
approximately 5%. Electron microscopy confirmed the presence of
abundant mitochondria in the cytoplasm of tumour cells. Although
patchy chromogranin positivity was unusual neurosecretory gran-
ules were not identified. The morphological features in conjunction
with immunohistochemistry and electron microscopy were typical
for an adrenocortical oncocytoma.

4. Discussion

Oncocytomas can occur in various organs, especially the kidneys
where oncocytomas represent around 3–7% of all renal neoplasms.2
They are much rarer in the endocrine, respiratory and gastrointesti-
nal tissues. They are predominantly benign tumours which may
reach considerable size if unrectected. However, there is little data
on the natural history of oncocytomas arising from the respective
organs with scattered reports of metastatic oncocytomas in the
literature.

Oncocytomas of the adrenal gland are particularly rare, although
exact overall incidence is unknown. They have been reported in a
wide age range between 15 and 77 years. They have been observed
to occur more frequently in females (2.5:1) and to congregate on the
left side (3.5:1).2 There are no identified environmental or genetic
risk factors. They are usually identified incidentally and the major-
ity are non-functioning. Fewer than 10 cases have been reported
of hormone secreting adrenal oncocytomas resulting in virilisation
and Cushing’s syndrome.3
The imaging characteristics of adrenal oncocytomas have been previously discussed by Shah et al.4 Notably, on CT imaging they demonstrate heterogeneous contrast enhancement with areas of hypodensity and fibrous encapsulation. These features are consistent for both benign and malignant histologies. Furthermore, the CT features do not distinguish adrenal oncocytomas from more common adrenocortical carcinoma. These findings are thus non-specific and offer no prognostic information. The diagnostic option of fine needle aspiration has been investigated by Quayle et al.5 They found that histological assessment of incidental adrenal lesions by fine needle aspiration was often unsuccessful in providing a sample of diagnostic value, had a high complication rate (notwithstanding the risks of needle track tumour seeding) and ultimately did not alter the clinical management. Surgical excision followed by a full histopathological assessment is thus best practise for all suspicious lesions. In our case, the MRI scan was particularly useful in delineating the lesion as arising from the adrenal gland.

Several efforts have been made to characterise oncocytomas based on their histological characteristics. Histological scoring systems have been proposed by Weiss for adenocortical tumours and more recently by Bisceglia et al. specifically with regards to oncocytomas to establish the histological grade.6,7 Bisceglia proposed major and minor criteria as part of a malignancy scoring system. The three major criteria are; high mitotic rate (>5 mitoses per 50 high power fields), atypical mitoses or venous invasion. The four minor criteria are; tumour size >10 cm or >200 g, tumour necrosis, capsular or sinusoidal invasion. Benign lesions would lack all of these features while the presence of any one or more major criteria categorises the lesion as a malignant oncocytoma. The presence of at least one major criterion yields a diagnosis of “uncertain malignant potential/borderline malignant”. On the basis of this Bisceglia scoring system, 53% of 53 reported cases of oncocytoma can be classed as benign, 19% as malignant and the remaining 28% as borderline/uncertain.2 The tumour in our patient was classified with “borderline” malignant potential and there were no other adverse features such as vascular or capsular invasion.

However, there remains uncertainty over the validity of such scoring systems and their ultimate clinical and prognostic significance. This appears to be due to small numbers and the limited long term follow up data for excised oncocytic lesions and their natural history. This makes the post operative surveillance regime difficult to rationalise. Of the 22 reported benign oncocytomas with long term follow up there was no documented recurrence after resection (follow up ranging 6–96 months; mean 34 months).7,8 In 6 cases of oncocytomas with borderline/uncertain malignant potential the disease-free follow up extends 10–61 months (mean 25.5 months). With limited data, there does not appear to be any instances of recurrence of benign or borderline oncocytomas following excision. In 9 malignant cases with reported follow up 5 were reported to be disease-free with a median reported follow up of 6 months (range 5–32 months) while 4 had developed recurrence.7,9,10 Interestingly in a detailed study of four malignant oncocytomas Hoang et al. concluded that no cytological or mitotic criteria can accurately predict biological behaviour of the tumour.9 They conclude that only tumour size, capsule/vessel invasion, necrosis and metastasis are reliable indicators of malignant disease.

Surgical excision is curative in the vast majority of adrenal oncocytomas. This may be carried out as an open procedure as in our case or via laparoscopic route for smaller lesions.11 Although there is no established role for chemotherapy or radiotherapy Juliano et al. report the use of empirical chemotheraphy and radiotherapy in a 15 year period of follow up of a single patient with metastatic adrenal oncocytoma.10 The initial reported histology in that patient after excision was that of a low malignant potential adrenocortical oncocyto. Three years later metastases in bone and liver were treated with resection followed by six cycles of chemotherapy. After a further six years a local recurrence at the adrenal bed was treated with radiotherapy.

In conclusion, adrenal oncocytoma is an extremely rare tumour with uncertain malignant potential. We advocate the use of MRI scan for delineation of the tumour characteristics, particularly in case of large right sided lesions. Surgical resection remains the mainstay of the treatment. Although scoring systems exist in order to prognosticate the tumour behaviour, several uncertainties preclude accurate prediction of malignant behaviour. Hence, a long term surveillance policy after surgical resection is advocated.

Conflict of interest statement

No potential conflicts of interests are known for this case report.

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Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

References

1. Peppa M, Karamitopoulou E, Nikolopoulos P, Peros G, Economopoulos T, Sotiriou A et al. Large adrenal oncocytoma with uncertain malignant potential: A case report and review of the literature. Endo-Prac 2010;1–14.
2. Dechter CB, Bostwick DG, Blute ML, Bryant SC, Zincke H. Renal oncocytoma: multifocality, bilateralism, metachronous tumor development and coexistent renal cell carcinoma. J Urol 1999;162(1):40–2.
3. Geraisizadeh B, Norouzzadeh B, Bolandparvaz S, Seifdahkt S. Functioning adrenocortical oncocyto: a case report and review of literature. Indian Pathol Microbiol 2008;51:237–9.
4. Shah RK, Oro A, Ozkan OS, Ernst RD, Hernandez JA, Chaudhary HB, et al. Adrenal Oncocyto: US and CT findings. JBR-BTR 2004;87:180–2.
5. Quayle FJ, Spiteri JA, Pierce RA, Lairmore TC, Moley JF, Brunt LM. Needle biopsy of incidentally discovered adrenal masses is rarely informative and potentially hazardous. Surgery 2007;142:497–502.
6. Weiss LM. Comparative histologic study of 43 metastasizing and nonmetasta-
sizing adrenocortical tumours. Am J Surg Pathol 1984;8:163–9.
7. Bisceglia N, Ludovico O, Di Mattia A, Ben-Dor D, Sandbank J, Pasquinielli G, et al. Adrenocortical oncocytic tumours: report of 10 cases and review of the literature. Int J Surg Pathol 2004;12:231–1231.
8. Lin B, Bonsib S, Mierau G, Weiss L, Medeiros LJ. Oncocytic adrenocortical neo-
plasms. Am J Surg Pathol 1998;22:603–14.
9. Hoang MP, Ayala AG, Alberos-Saavedra J. Oncocytic adrenocortical carcinoma: a morphologic, Immunohistochemical and ultrastruc-
tural study of four cases. Mod Pathol 2002;15(9):973–8.
10. Juliano J, Cody RI, Suh JH. Metastatic adrenocortical oncocytoma: a case report. Urol Oncol 2008;26(2):198–201.
11. Eldahshan S, Zeccolini G, Guerini A, Breda G. Laparoscopic transperineal adrenalectomy for adrenocortical oncocytoma. Arch Ital Urol Androl 2008;80(2):82–4.