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Case Report

Adrenal Oncocytoma: A Case Report and Review of the Literature

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Abstract

Adrenal oncocytomas are one of the rarest differentials of an adrenal "incidentaloma", an adrenal mass typically found unexpectedly on radiological examination. An adrenal incidentaloma > 4 cm is likely to be malignant; however adrenal oncocytomas are large, averaging 8 cm, and very rarely demonstrate malignant potential. They are typically rounded lesions, encapsulated and well circumscribed. Metabolic activity is rare, although there are reports of patients presenting with features of Cushing's disease or Phaeochromocytoma. There are unclarified features of an adrenal oncocytoma on CT, making radiological diagnosis difficult. MRI can be useful in confirming the lesion as arising from the adrenal gland. Management should involve surgical resection, laparoscopic adrenalectomy being the gold standard approach. A diagnosis of adrenal oncocytoma is based on the classical histological and immunochemistry findings, although it can remain difficult to predict metastatic behaviour.

Keywords: *Urology; Adrenal Gland; Benign; Oncocytoma; Adrenal Incidentaloma*

Introduction

Oncocytomas are epithelial tumours composed of cells with abundant eosinophilic granular cytoplasm packed with mitochondria [1]. Oncocytomas can occur in various organs such as the kidney, thyroid, pituitary gland and parathyroid glands. They have also been reported in the respiratory and gastrointestinal tracts [2]. They are particular prevalent in the kidney, representing around 3 - 7% of all renal neoplasms [3].

Case Report

A 46-year-old gentleman presented to the Urology team having been referred by the Dermatology department, known to them with a long history of psoriasis. He was currently on Methotrexate and had never required any biological agents. He was otherwise fit and well, although had also recently been referred to the Rheumatologists due to worsening pain in his elbows, shoulder and DIP joints. He was a cigarette smoker.

The Dermatology team had arranged an ultrasound scan of his liver due to newly deranged LFTs. This showed a normal liver but the incidental finding of a well-defined, solid left adrenal mass measuring 5 cm. CT chest, abdomen and pelvis confirmed the adrenal mass, with no evidence of any primary disease elsewhere, including a lung malignancy. The patient was discussed at a multidisciplinary meeting and an interval, focussed adrenal CT scan was arranged for further characterisation (Figure 1). Following review of this scan it was felt the adrenal mass was increasing in size and was deemed to be indeterminate and not definitely an adenoma - absolute washout of contrast was 52.2% with relative washout 27.5%. A Nephrologist was consulted, who arranged for two 24-hour urine results. These showed normal levels of metanephrines, rendering the diagnosis of phaeochromocytoma highly unlikely.

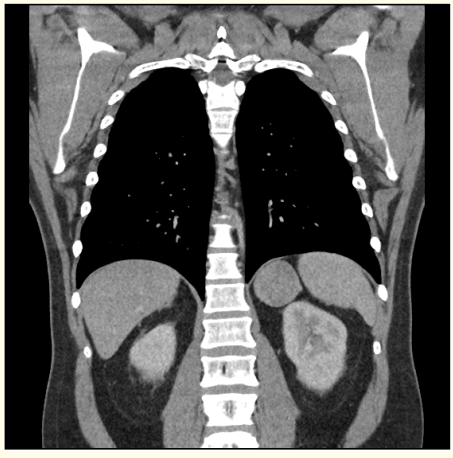

Figure 1: *CT adrenal characterisation scan revealing a 4.8cm mass arising from the adrenal gland.*

Following further multidisciplinary discussion, a consensus was reached that excision rather than biopsy would be the best course of treatment. It was explained to the patient that we were unsure of its nature, but without removal we could not be sure whether it was a cancer or not.

A laparoscopic left adrenalectomy was performed. The operation was uncomplicated and the patient discharged the next day. The specimen underwent full histological examination. Macroscopically the specimen was described as a smooth yellow nodule 45mm in diameter with attached fatty tissue 65 x 40 x 20 mm including unremarkable background adrenal gland. Soft and haemorrhagic areas present in the nodules on slicing. Microscopically, the background adrenal gland was within normal limits. There was a well circumscribed tumour arising from the gland composed almost exclusively of large cells with intensely eosinophilic dense cytoplasm. There was moderate nuclear atypia with a few multinucleate cells. No significant clear cell population was present. There was no evidence of fibrosis, confluent necrosis, lymphovascular, sinusoidal or capsular invasion by tumour. No mitotic activity identified in 50 HPF. The features were those of a benign adrenal oncocytoma (Figure 2 and 3).

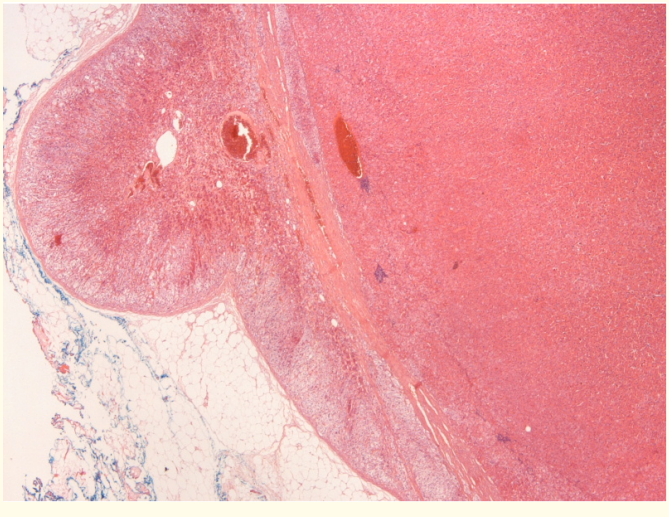

Figure 2: *20x magnification revealing a well circumscribed tumour and adjacent normal adrenal gland.*

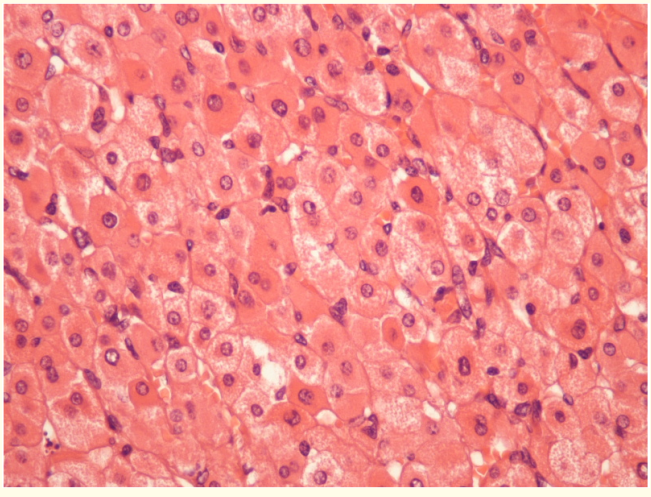

Figure 3: *400x magnification of oncocytoma, revealing large cells with intensively eosinophilic dense cytoplasm.*

The patient was informed of the diagnosis and continued to make a good recovery. It was explained that this was a rare, benign tumour. A surveillance CT scan was booked in a years time.

Discussion

Oncocytomas are epithelial tumours composed of cells with abundant eosinophilic granular cytoplasm packed with mitochondria [1]. Oncocytomas can occur in various organs such as the kidney, thyroid, pituitary gland and parathyroid glands. They have also been reported in the respiratory and gastrointestinal tracts [2]. They are particular prevalent in the kidney, representing around 3 - 7% of all renal neoplasms [3].

Oncocytomas of the adrenal gland, however, are incredibly rare. Having first been reported by Kakimoto in 1986, only 120 cases of adrenal oncocytoma have been reported [4]. Adrenal oncocytomas are typically discovered incidentally. They are therefore a very rare differential diagnosis of an adrenal 'incidentaloma" - an adrenal mass, typically one cm or more in diameter, that is discovered by chance during a radiological examination performed for indications other than evaluation of adrenal disease [1]. Others differentials include cortical adenoma, phaeochromocytoma, adrenocortical carcinoma, granulomas, adrenal cyst, myelolipoma, ganglioneuroma and metastatic deposits [1].

Adrenal oncocytomas are typically non-functioning, benign tumours which vary in size but are generally larger than 6 cm. Interestingly, adrenal incidentalomas greater than 4 cm in size are likely to be malignant; adrenal oncocytomas are therefore an exception to this rule. Slightly more common in females, adrenal oncocytomas have been reported in a wide age group ranging from 15 to 77 years [3]. No predisposing risk factors have been identified (genetic or environmental) [5]. The vast majority of adrenal oncocytomas described in the literature are non-functioning. However, recent studies indicate metabolic activity may be present in up to 10-20% of cases. An adrenal oncocytoma occurring with a Cushing's syndrome, phaeochromocytoma or aldosteronoma have all been described [2]. As was performed in this case study, careful biochemical analyses should be performed to exclude a functioning tumour, in particular a phaeochromocytoma, as this will affect pre- and post-operative planning.

Fat concentration is a useful differentiator of benign and malignant adrenal lesions - malignant lesions are generally lipid-poor, whereas the vast majority of benign lesions, such as adrenocortical adenomas, are lipid-rich with a lower attenuation on CT scan (10 HU or less) [5]. Adrenal oncocytomas, despite being benign lesions, have been found to have attenuation ranging from 20 to 40 HU [2]. Furthermore, the features adrenal oncocytomas display often replicate those of a malignant process. For example, fibrous encapsulation and heterogeneous contrast enhancement are typically present, findings also present in some malignant tumours [3]. Features which would more likely support a diagnosis of the malignant adrenocortical carcinoma, such as tissue invasion of fat planes and vascular structures, are

absent and must be excluded [3]. MRI has been used in some instances and is useful at demonstrating the mass to be arising from the adrenal as well as differentiating adrenal adenomas from other disease. It is unable to detect adrenal oncocytomas, however.

Unfortunately, given the limitations described, it is difficult to diagnose adrenal oncocytoma radiologically - both CT and MRI are unable to detect an adrenal oncocytic neoplasm or differentiate an oncocytoma from other malignant lesions [5]. Although occasionally reported in the literature, the role of fine needle aspiration (FNA) pre-operatively to obtain a diagnosis is debatable, with a sample often deemed to be insufficient. Moreover, FNA comes with the additional risk of seeding malignant cells if the tumour turns out to be a sinister process [3].

Given the diagnostic limitations, surgical excision is the management of choice for any suspicious adrenal lesion. The gold standard approach for a small adrenal tumour is a laparoscopic adrenalectomy, providing less intraoperative bleeding, a faster recovery and shorter hospital stay than open techniques. For larger adrenal lesions, such as an adrenal oncocytoma, laparoscopic adrenalectomy is deemed to be safe, so long as the pre-operative imaging has displayed a well encapsulated tumour with no evidence of local invasion and or regional lymph node involvement [5].

On macroscopic histological examination, adrenal oncocytomas appear round, well circumscribed and encapsulated [1]. On average they have a diameter of 8 cm. They are typically dark brown in colour with areas of haemorrhage and necrosis on cut section [5]. On microscopic histological analysis the cells may be arranged in solid, trabecular tubular or papillary patterns. The cells themselves are the same size, highly eosinophilic and granular. Under the electron microscope, as is typical of adrenocortical cells, the oncocytic cells contain abundant mitochondria [5].

As has been discussed, the great majority of adrenal oncocytomas are benign. However, it has been reported that up to 22% of tumours have shown malignant potential [2]. Malignant oncocytomas are very rare and have a poor prognosis [1]. The Weiss system was introduced to differentiate between a benign and malignant neoplasm, however this system was deemed unreliable for the adrenal gland. In more recent years, Bisceglia., *et al.* proposed a malignancy scoring system of major and minor criteria, specific to oncocytomas, to establish the histological grade and malignancy potential. The three major criteria are as follows: Mitosis count > 5/50 HPF, atypical mitosis and venous invasion, with four minor criteria: tumour diameter > 10 cm and/or weight > 200g, necrosis, capsular invasion and sinusoidal infiltration. A benign oncocytoma would reveal none of these features on histological examination. The presence of a single major criteria would categorise the lesion as malignant. Having at least one minor criterion would lead to a diagnosis of uncertain malignant potential (borderline) [3,5].

Despite these scoring systems there is limited long term follow up data for resected oncocytic lesions. Determining an appropriate post-operative surveillance strategy is therefore not without difficulty. With the limited data available, there have been no reports of recurrence of benign or borderline oncocytomas following excision. In 9 malignant cases followed 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, demonstrating the poor prognosis of such tumours [3]. With the absence of clear, objective guidance on how to review these patients post-operatively, Mearini., *et al.* recommended a regular follow up for a minimum of 5 years [5].

Conclusion

In conclusion, adrenal oncocytomas are very rare adrenal incidentalomas that are typically large, non-functioning and benign. CT and MRI scans fall short of providing a pre-operative diagnosis, rendering surgical excision the mainstay of treatment, ideally via a laparoscopic approach. Diagnosis is based on classical histological findings of an oncocytoma. One must be aware of the malignant potential of these lesions and histological features which would suggest such a diagnosis. There are no clear guidelines on how one should follow up these patients, however given the uncertainties surrounding the malignant potential of these lesions, it seems sensible to implement a long-term surveillance policy.

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