CASE REPORT

Imaging appearance of bulk fat within an oncocytic adrenocortical neoplasm, a rare and potentially malignant tumour

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ABSTRACT. Oncocytic adrenocortical neoplasm is a rare adrenal tumour that usually follows a benign clinical course. In some cases, however, these tumours have exhibited malignant behaviour. Here, we present the first published case showing bulk fat within an oncocytic adrenocortical neoplasm on CT and MRI, a finding that mimics fat within an adrenal myelolipoma. The distinction between these entities is important, as the current suggested management of an oncocytic adrenocortical neoplasm is resection with subsequent imaging surveillance.

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Incidental adrenal lesions are common and are estimated to occur in about 4% of abdominal CT examinations [1]. The presence of even a small amount of fat within an adrenal lesion is often deemed a specific finding for establishing a diagnosis of a benign myelolipoma [2, 3]. In this report we present the CT and MRI findings of an incidentally discovered adrenal lesion containing bulk fat that proved to be an oncocytic adrenocortical neoplasm. The biological behaviour of these rare lesions is not yet fully understood and resection and imaging surveillance are recommended. To our knowledge, the imaging appearance of an oncocytic adrenocortical neoplasm containing fat has not been previously demonstrated.

Case

A 52-year-old male underwent CT colonography after an incomplete conventional colonoscopy. CT showed no evidence of colonic neoplasm; however, there was a 7.6 × 7.4 cm well-circumscribed heterogeneous left adrenal mass that contained several scattered areas of bulk fat (Figure 1a). The interpreting radiologist favoured a diagnosis of myelolipoma, but suggested short-term imaging follow-up or biopsy be performed given its large size. MRI obtained 1 month after the initial CT examination demonstrated a heterogeneously enhancing mass (Figure 1b) containing areas of fat, as demonstrated by areas of high signal intensity on in-phase T_1 weighted images (Figure 1c) with a peripheral rim of signal drop-out along its border with the adjacent kidney on out-of-phase T_1 weighted images (Figure 1d) and diffuse loss of signal on chemically selective fat-suppressed T_1 weighted images (Figure 1e). MRI obtained 3 months and CT

obtained 5 months after the initial CT showed overall stable size and appearance of the left adrenal lesion; no new abnormality was identified. The patient elected at this time for resection of the lesion, given its size and continued uncertainty regarding the underlying diagnosis. Laparoscopic left adrenalectomy was performed. On gross examination, there was a circumscribed nodular mass that measured 8.5 × 6.0 cm and weighed 182 g. Histological evaluation of the mass revealed tumour cells arranged in solid sheets and demonstrating abundant eosinophilic granular cytoplasm (Figure 2). There was no identified area of tumour necrosis, atypical mitoses or elevated mitotic activity (proliferation index by MIB-1 <2%). The resection margins were negative and no vascular invasion was identified. Scattered areas of benign, mature adipose tissue were noted within the mass (Figure 2). Immunohistochemical staining showed tumour cells to be positive for vimentin, calretinin and melan-A, weakly positive for synaptophysin and negative for cytokeratin (Cam5.2 and AE1/AE3), chromogranin and S-100. Given these features, the lesion was diagnosed as an oncocytic adrenocortical neoplasm with benign features. The patient has undergone continued imaging surveillance; most recent follow-up CT and MRI performed 3 years post-operatively showed no evidence of recurrent disease.

Discussion

Oncocytic adrenocortical neoplasm is an adrenal tumour that consists entirely or predominantly of oncocytes, large polygonal epithelial cells filled with eosinophilic granules that correspond with densely packed mitochondria on electron microscopy [4–9]. The lesion is exceedingly rare with fewer than 80 cases having been reported since the entity was first described in 1986 [10]. Although Lin et al [11] described the “presence of discrete, often relatively large foci of

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Figure 1. A 52-year-old male with left adrenal mass containing bulk fat, diagnosed as an oncocytic adrenal neoplasm. (a) A non-enhanced axial CT image demonstrates a large left adrenal mass containing a small area of bulk fat (arrow). Additional areas of fat with a similar appearance were scattered throughout the remainder of the mass. (b) Axial contrast-enhanced 3D gradient-echo T_1 weighted image demonstrates a large left adrenal mass with extensive heterogeneous areas of enhancement. In-phase T_1 weighted image (c, arrow) demonstrates T_1 hyperintensity corresponding with an area of fat on CT. This area demonstrates a peripheral rim of signal drop-out along its border with the adjacent kidney on the corresponding out-of-phase T_1 weighted image (d, arrow). In addition, this region shows diffuse loss of signal on the chemically selective fat-suppressed T_1 weighted image (e, arrow). These signal characteristics confirm the presence of bulk fat within the lesion.

Figure 2. A 52-year-old male with left adrenal mass containing bulk fat, diagnosed as an oncocytic adrenal neoplasm. A photomicrograph of the left adrenal mass (haematoxylin and eosin stain $\times 20$) demonstrates solid sheets of tumour cells with abundant eosinophilic granular cytoplasm, compatible with an oncocytic adrenocortical neoplasm (solid black arrow). There are areas of adipose tissue within the mass (dashed black arrow). The normal adrenal cortex from which the tumour is arising is also shown (open white arrow).

adipose tissue" in three oncocytic adrenocortical neoplasms, showing a photomicrograph of the histological appearance of such adipose tissue in one case, our report is the first to demonstrate the radiological appearance of bulk fat within an oncocytic adrenocortical neoplasm on CT or MRI.

The lesion most commonly demonstrates a benign histological appearance and biological behaviour. In such cases, the lesion has been designated by some authors as an adrenocortical oncocytoma [4, 5, 9, 11], analogous to the oncocytoma that is recognised in the kidney and salivary, thyroid and parathyroid glands [7]. However, oncocytic adrenocortical neoplasms have also presented in some cases as carcinomas with a malignant clinical course; for example, demonstrating invasion of the inferior vena cava [8, 12–14], local recurrence following resection [15, 16] and bone, liver and lung metastases [15], with death as a reported outcome [17]. Thus, the major challenge in the management of these neoplasms is to distinguish benign from malignant lesions. Yet, the entity remains incompletely understood, with no single clinical, histological or immunochemical feature able to predict the clinical course of an individual lesion. To assist in their categorisation, Bisceglia et al. [17] proposed a classification system that divided oncocytic adrenocortical neoplasms into three broad groups on the basis of histological features with different prognostic implications; however, given the small number of lesions reported, as well as the short clinical follow-up available for most lesions, this classification system is considered controversial [15] and caution is still warranted when applying histological criteria to individual lesions [17]. Indeed, some reported cases in the literature have demonstrated a more aggressive clinical course than would be predicted using this classification system [8, 12, 13, 17]. Until these lesions are better understood, long-term follow-up of oncocytic adrenocortical neoplasms is suggested [4, 5].

The adrenal lesion in the case reported here was initially thought likely to represent a myelolipoma, given the presence of scattered areas of fat. However, the radiological detection of fat has been described within other adrenal lesions, including neoplasms such as liposarcoma and teratoma [18]. Numerous authors have suggested a role for tissue sampling when an adrenal mass is considered suspicious but indeterminate for myelolipoma [18–21]. The large size, heterogeneity and predominantly solid nature of the lesion in this case raised sufficient concern that the lesion was not in fact a myelolipoma and tissue diagnosis was suggested. Krishnamurthy et al [8] diagnosed an indeterminate adrenal lesion as an oncocytic adrenocortical neoplasm using fine-needle aspiration (FNA), noting that oncocytic cells had a characteristic appearance that was easily recognised in the FNA smear and allowed for a definite diagnosis to be made using this technique. Although FNA might correctly diagnose an indeterminate adrenal lesion as an oncocytic adrenocortical neoplasm, the diagnosis will not alter the need for surgical management [4, 15]; it is not possible to assess for capsular or vascular invasion based on cytology alone and focal areas of abnormal mitotic activity or necrosis might be missed owing to sampling error [17]. The difficulty of attempting to classify oncocytic adrenal

neoplasms on the basis of a limited tissue sample is demonstrated by a case reported in the literature in which recurrent neoplastic disease resulted in death. In this instance the neoplasm was initially classified as benign, but was subsequently reclassified as malignant when additional slides from the original tumour were reviewed and found to demonstrate high mitotic activity [17].

In summary, this is the first report of fat within an oncocytic adrenocortical neoplasm demonstrated on CT and MRI. Despite being uncommon, this diagnosis must at least be considered in the presence of a large adrenal mass demonstrating predominantly soft tissue with only scattered areas of fat. In these cases biopsy can be performed, which may readily establish the diagnosis of oncocytic adrenocortical neoplasm. Unlike myelolipoma, resection and continued surveillance are warranted once the diagnosis of oncocytic adrenocortical neoplasm is established, given the evolving understanding of its biological behaviour.

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