CASE REPORT

Unilateral adrenal mass as the sole initial manifestation of differentiated thyroid cancer

Geethalakshmi Sampathkumar,¹ Arun S Menon,¹ Bindhu M R,² Vasantha Nair¹

¹Department of Endocrinology and Metabolism, Amrita Institute of Medical Sciences and Research Centre, Cochin, Kerala, India

²Department of Pathology, Amrita Institute of Medical Sciences and Research Centre, Cochin, Kerala, India

Correspondence to

Dr Geethalakshmi Sampathkumar, drgeethalakshmi86@gmail.com

Accepted 3 September 2017

SUMMARY

Unilateral adrenal metastases without disseminated disease has rarely been reported in differentiated thyroid carcinoma (DTC). A 72-year-old female presented with vague abdominal discomfort and loss of appetite of 2 months duration. She had undergone left hemithyroidectomy for a benign thyroid nodule 18 years ago. A contrast CT of the abdomen showed a large left adrenal mass measuring 11×9 cm, suspicious of adrenocortical carcinoma. Hormonal evaluation was in keeping with a non-functional tumour. The patient underwent left adrenalectomy, histopathology of which revealed metastatic well-differentiated thyroid carcinoma. Ultrasound of thyroid done postoperatively showed a subcentimetric hypoechoic lesion with increased vascularity and microcalcifications in the right thyroid bed. Histopathology from a completion thyroidectomy specimen was consistent with follicular variant of papillary thyroid carcinoma. She was treated with high-dose radioiodine ablation therapy and has remained disease-free on follow-up for more than a year.

BACKGROUND

Unilateral adrenal metastases as an initial presentation of differentiated thyroid carcinoma (DTC) and without disseminated disease is very uncommon. Our case highlights the importance of considering DTC in the differential diagnosis of isolated adrenal metastases.

CASE PRESENTATION

A 72-year-old female with a background history of systemic hypertension and bronchial asthma presented with vague abdominal discomfort and loss of appetite of 2 months duration. A routine ultrasound done in local hospital revealed a left-sided suprarenal mass. Hence, she was referred to us for further evaluation. She had a history of left hemithyroidectomy 18 years ago for benign thyroid nodule and has been on levothyroxine replacement since then. However, no records of the surgery or histopathology were available. She denied any history of episodic palpitation, sweating or headache. Examination of neck revealed a faint scar over the front of neck but no palpable thyroid nodule. Her blood pressure was within normal limits with no postural drop. She had no cushingoid features or signs of virilisation. CT abdomen with contrast demonstrated a 11×9 cm heterogenous enhancing mass lesion noted in left suprarenal region displacing left kidney and renal vein suggestive of adrenocortical carcinoma (figure 1).

INVESTIGATIONS

Hormonal evaluation performed were within normal limits. Her serum cortisol was 235 nmol/L (102–535 nmol/L), plasma metanephrines 106.47 pmol/L (<456.3 pmol/L), normetanephrines 226 pmol/L (<982.8 pmol/L), dehydroxy epianandrosterone sulfate 1.88 micromol/L (2.71–11.38 micromol/L) and testosterone 1.4 nmol/L (0.4–2 nmol/L). Low-dose dexamethasone suppression test with 1 mg dexamethasone orally showed adequate suppression of serum cortisol (6 nmol/L).

DIFFERENTIAL DIAGNOSIS

Adrenocortical carcinoma was our primary differential diagnosis based on unilaterality, large size and heterogeneity of the mass on CT scan. Although adrenal secondaries was considered, there was no evidence of a primary focus at that stage.

TREATMENT

She underwent left laparoscopic adrenalectomy and a 12×9×8.5 cm nodular well-encapsulated lesion showing normal adrenal tissue in periphery was removed. Histopathology was consistent with (thyroglobulin positive)metastatic well-differentiated follicular variant of papillary thyroid carcinoma (figures 2 and 3). Postoperatively, an ultrasound of the thyroid gland showed a suspicious 7×7 mm hypoechoic nodule with internal vascularity and

Figure 1 CT of abdomen showing the large heterodense left adrenal mass.

CrossMark

To cite: Sampathkumar G, Menon AS, M R B, et al. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-221296

Figure 2 Histopathology of left adrenal showing the presence of metastatic thyroid carcinoma in the background of normal adrenal tissue.

microcalcification in the right lobe. Although fine-needle aspiration cytology was non-diagnostic, in view of the strong clinical suspicion, she underwent completion thyroidectomy. Histopathology confirmed 1×2cm infiltrating multifocal papillary thyroid carcinoma—follicular variant ([figure 4](#)). Immunohistochemistry showed patchy positivity for cytokeratin 19, Galectin 3, Hecto Battifora Mesothelial 1 and CD56. Diagnostic whole body I-131 scintigraphy revealed minimal residual thyroid tissue with no uptake in adrenal region or elsewhere. Her stimulated thyroglobulin was 4.78 mcg/L. She was ablated with high-dose radioiodine (4736 MBq I-131).

OUTCOME AND FOLLOW-UP

She has remained disease-free on follow-up 12 months after ablative radioiodine therapy, with repeat whole body I-131 scintigraphy showing no uptake ([figure 5](#)) and an undetectable stimulated thyroglobulin level (<0.04 mcg/L).

Figure 3 High-power image of the left adrenal showing characteristic nuclear features of papillary thyroid carcinoma with follicular arrangement.

Figure 4 Histopathology of thyroid tissue showing follicular variant of papillary thyroid carcinoma.

DISCUSSION

Thyroid cancer is the most commonly encountered endocrine malignancy with an increasing incidence over the past few decades. Well-differentiated thyroid malignancies constitute 90% of these cases and usually run an indolent clinical course with excellent prognosis.¹ Distant metastases from DTC is uncommon with frequencies reported to be less than 13%.² It is even less common for DTC to have distant metastases at presentation (1%–4%). More than 50% of patients with DTC with distant metastases die from the disease during follow-up.³ Most of them present with regional lymph nodal metastasis followed by lung and bone. Very rarely they can metastasise to unusual sites like the breasts, skin, eyes, pancreas, liver, kidney and adrenal glands. Almost always, these rare presentations are associated with advanced disease and poor prognosis.⁴ Prognosis of patients with metastatic thyroid cancer is usually based on age, tumour size, grade, subtype and metastatic site.⁵

Thyroid cancer metastasising to adrenal gland is quite rare. Around 31 cases of adrenal metastasis have been reported so far in literature. Almost all those patients had advanced disseminated disease.⁶ To our knowledge, only one case has been reported so far with adrenal metastasis as the presenting feature. However, that patient had a synchronous renal and adrenal metastasis from a papillary thyroid cancer with loco regional disease.⁷ Our patient only had unilateral adrenal metastases at presentation with no further evidence of metastatic disease on follow-up to a year. Although adrenal metastases have been reported from all subtypes of thyroid cancer, majority of them were from papillary and follicular carcinoma.⁸

Histopathology revealed infiltrative follicular variant of papillary thyroid cancer (FVPTC) in our patient. FVPTC has an incidence ranging between 18% and 25% from the Western world and a very low incidence of 1.5% in Asian countries. The behaviour of FVPTC depends on whether it is infiltrative or encapsulated. The infiltrative tumours were found to have more extrathyroidal extension and nodal metastases. On the other hand, the encapsulated FVPTC especially with capsular or vascular invasion behave like follicular carcinoma of thyroid.^{9 10}

Usually, adrenal metastases from DTC are asymptomatic and rarely iodine avid. They are most often diagnosed by other imaging modalities such as positron emission tomography CT/single-photon emission CT.⁴ Once diagnosed, adrenal metastases

Figure 5 Iodine 131 whole body scan 6 months after thyroidectomy showing no abnormal uptake in thyroid or adrenal gland.

from thyroid are managed by adrenalectomy followed by radioiodine therapy.¹¹ In general, prognosis is poor in these patients as they are most often associated with advanced disease, unlike our patient whose initial presentation was as a large isolated adrenal mass with no other evidence of metastasis.

We report this case for the following reasons: the (1) rarity of adrenal metastases at presentation in DTC, (2) solitary large adrenal deposit with no evidence of metastases elsewhere on radioiodine scan and (3) the absence of any evidence of recurrence after a year of follow-up.

Learning points

- ▶ Although differentiated thyroid carcinoma (DTC) usually behaves in an indolent manner, occasionally they can present with distant metastases.
- ▶ Although metastases at presentation in DTC is usually associated with disseminated disease, isolated distant metastases is possible and could have a good prognosis.
- ▶ This case also highlights the need for a more thorough search for a primary focus in all cases of adrenal mass lesions.

Contributors GS and BMR contributed to the conception, design, analysis and interpretation of the data. ASM and VN were responsible for the drafting the article and the final approval of the version to be published. ASM was involved in revising the manuscript critically for important intellectual content. All authors read and approved the final manuscript.

Disclaimer I, GS, on behalf of all authors confirm that this work is original and also verify that all the authors have made a significant contribution to the findings in this case report. I certify that neither this manuscript nor one with substantially similar content under my authorship has been published or is being considered for publication elsewhere.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

© BMJ Publishing Group Ltd (unless otherwise stated in the text of the article) 2017. All rights reserved. No commercial use is permitted unless otherwise expressly granted.

REFERENCES

- 1 Blumhardt R, Wolin EA, Phillips WT, *et al.* Current controversies in the initial post-surgical radioactive iodine therapy for thyroid cancer: a narrative review. *Endocr Relat Cancer* 2014;21:R473–R484.
- 2 Xu B, Tuttle RM, Sabra MM, *et al.* Primary thyroid carcinoma with low-risk histology and distant metastases: clinicopathologic and molecular characteristics. *Thyroid* 2017;27:632–40.
- 3 See A, Iyer NG, Tan NC, *et al.* Distant metastasis as the sole initial manifestation of well-differentiated thyroid carcinoma. *Eur Arch Otorhinolaryngol* 2017;274.
- 4 Song HJ, Xue YL, Xu YH, *et al.* Rare metastases of differentiated thyroid carcinoma: pictorial review. *Endocr Relat Cancer* 2011;18:165–74.
- 5 Kumar A, Nadig M, Patra V, *et al.* Adrenal and renal metastases from follicular thyroid cancer. *Br J Radiol* 2005;78:1038–41.
- 6 Khorjekar RG, Klubo-Gwiedzinska J, Van Nostrand D, *et al.* In: *Summary of rare sites of metastasis secondary to differentiated thyroid cancer. A comprehensive guide to clinical management*, 3rd edn. New York, USA: Springer Publishing, 2016:735–44.
- 7 Ginzburg S, Reddy M, Veloski C, *et al.* Papillary thyroid carcinoma metastases presenting as ipsilateral adrenal mass and renal cyst. *Urol Case Rep* 2015;3:221–2.
- 8 Chentli F, Bendali M, Boutekedjiret F, *et al.* Adrenal metastases from thyroid cancers: three personal cases and literature review. *J Endocrinol Diabetes Mellit* 2015;3:11–15.
- 9 Daniels GH. Follicular variant of papillary thyroid carcinoma: hybrid or mixture? *Thyroid* 2016;26:872–4.
- 10 Bychkov A, Hirokawa M, Jung CK, *et al.* Low rate of noninvasive follicular thyroid neoplasm with papillary-like nuclear features in Asian practice. *Thyroid* 2017;27:983–4.
- 11 Farina E, Monari F, Tallini G, *et al.* Unusual thyroid carcinoma metastases: a case series and literature review. *Endocr Pathol* 2016;27:55–64.

Copyright 2017 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow