CASE REPORT

Curative surgical management of isolated adrenal recurrence of oesophageal adenocarcinoma

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SUMMARY

Adrenal metastases of oesophageal adenocarcinoma are rarely detected in the clinical setting, more frequently being found as an incidental postmortem finding in the presence of widespread metastases. With improvements in the sensitivity of radiological diagnostic modalities, the incidence of adrenal tumour detection is on the rise. We report herein a particularly rare case of primary operative management by adrenalectomy for an isolated right-sided adrenal metastasis secondary to oesophageal adenocarcinoma, with a long-term survival.

BACKGROUND

This case represents a rare presentation of oesophageal cancer successfully managed through operative resection of the adrenal gland. The case highlights the unpredictable nature of oesophageal adenocarcinoma and the potentially curative outcome attainable after a long disease-free interval.

CASE PRESENTATION

A 50-year-old man presented with marked reflux symptoms, and endoscopy revealed a large ulcerated growth in the lower oesophagus, a Siewert type I adenocarcinoma of the oesophagogastric junction (AEG 1) (figure 1). Histological analysis confirmed the presence of moderately-to-poorly differentiated adenocarcinoma. CT and enhanced-ultrasound showed no evidence of lymphadenopathy or distant metastases, and he was clinically stage cT3N0. His medical history was notable for coronary artery disease, hypertension and morbid obesity (body mass index of 44.3 kg/m²). He had had a coronary artery bypass graft 11 years previously. He was a taxi driver and an ex-smoker.

Figure 1 Invasive moderately-to-poorly differentiated adenocarcinoma in the oesophagus.

Figure 2 Metastatic poorly differentiated adenocarcinoma in the adrenal gland.

Following a multidisciplinary team discussion it was considered that he should undergo a primary surgery rather than a multimodal approach, and he underwent a two-stage oesophagectomy 1 week later. Postoperative recovery was uncomplicated and the patient proceeded to standard outpatient follow-up as per the oesophageal cancer protocol in our institution.¹

INVESTIGATIONS

Four years after the initial diagnosis, routine surveillance CT revealed a 3.5 cm right adrenal mass suspicious for metastasis. Urinary catecholamine and metanephrine levels were normal, CT-guided biopsy was undertaken. Histopathology revealed an adenocarcinoma with similar cytonuclear morphology to the primary neoplasm, consistent with a metastasis from that site (figure 2). A CT positron emission tomography (PET) illustrated increased 2-fluoro-D-deoxyglucose (FDG) avidity within the right adrenal mass standard uptake value 10.9 and no sites of distant metastasis (figure 3). An endoscopy outruled the recurrence of oesophageal tumour.

DIFFERENTIAL DIAGNOSIS

Apart from metastatic disease, causes of focal adrenal uptake on PET/CT include benign and malignant pheochromocytoma, giant adrenal myelolipoma and adrenocortical carcinoma. Bilateral uptake, while worrying for metastasis, can also be due to adrenal hyperplasia or Cushing's syndrome.

TREATMENT

An open right adrenalectomy was undertaken with no postoperative complications.

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Figure 3 Fluoro-D-deoxyglucose avid mass in the right adrenal gland on positron emission tomography/CT.

OUTCOME AND FOLLOW-UP

The patient remains well and disease-free over 4 years postadrenalectomy, and 8 years following an oesophageal cancer resection.

DISCUSSION

Oesophageal carcinoma frequently presents with distal metastases, commonly found in the liver, lung and bone. The adrenal gland is the fourth commonest site of visceral metastasis secondary to oesophageal carcinoma but is usually diagnosed in the setting of widespread metastasis to multiple sites at postmortem. Metastatic infiltration of the adrenal gland is common, partly because of the rich sinusoidal blood supply and high local concentration of corticosteroids, which are thought to promote implantation. Studies indicate a rate of 6% and 10–13.6% from tumour registries and postmortem studies, respectively.^{2–4}

Adrenal metastases are increasingly detected during investigation of non-adrenal malignant disease due to improvements in imaging modalities.⁵ The discovery of adrenal metastasis in a patient with an operable primary tumour would traditionally have rendered the disease advanced and inoperable. A number of case reports have reported favourable outcomes and long-term survival in a variety of primaries including lung, melanoma, rectum, liver, kidney, thyroid, where synchronous or metachronous adrenal metastatic disease present.^{6–7} The sensitivity and specificity of PET/CT in characterising adrenal lesions in patients with known primary tumours are 100% and 94%,

respectively. The sensitivity of PET/CT is equal to MRI and superior to CT.⁸

Cases of isolated metastasis to the adrenal gland following oesophagectomy for oesophageal carcinoma managed successfully by adrenalectomy are rare. One report on adrenalectomy undertaken contemporaneously with oesophagectomy for a synchronous, isolated adrenal metastasis exists. This case highlights the unpredictable behaviour of oesophageal adenocarcinoma, and the curative potential of surgery in this rare scenario where the metastasis was isolated and there was a long disease-free interval since the primary curative therapy that may have portended favourable biology and outcome.

Learning points

- ▶ Oesophageal adenocarcinoma is an unpredictable malignancy and extended follow-up is crucial.
- ▶ While rare, oesophageal adenocarcinoma can result in adrenal metastases historically.
- ▶ In the setting of isolated disease, this is potentially curable.

Contributors JL identified the case. KO'S and AM gathered the case information and wrote the initial draft. JL and JVR edited the final draft prior to submission.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Reynolds JV, Donohoe CL, McGillicuddy E, *et al.* Evolving progress in oncologic and operative outcomes for esophageal and junctional cancer: lessons from the experience of a high-volume center. *J Thorac Cardiovasc Surg* 2012;143:1130–7e1.
- 2 Hess KR, Varadhachary GR, Taylor SH, *et al.* Metastatic patterns in adenocarcinoma. *Cancer* 2006;106:1624–33.
- 3 Lam KY, Lo CY. Metastatic tumours of the adrenal glands: a 30-year experience in a teaching hospital. *Clin Endocrinol* 2002;56:95–101.
- 4 Yoshizumi Y, Morisaki Y, Koike H, *et al.* Successful combined resection of carcinoma of the esophagus and adrenal metastasis: report of a case. *Surg Today* 1997;27:330–3.
- 5 Low G, Dhaliwayo H, Lomas DJ. Adrenal neoplasms. *Clin Radiol [Review]* 2012;67:988–1000.
- 6 Lo CY, van Heerden JA, Soreide JA, *et al.* Adrenalectomy for metastatic disease to the adrenal glands. *Br J Surg* 1996;83:528–31.
- 7 Twomey P, Montgomery C, Clark O. Successful treatment of adrenal metastases from large-cell carcinoma of the lung. *JAMA* 1982;248:581–3.
- 8 Yun M, Kim W, Alnafisi N, *et al.* 18F-FDG PET in characterizing adrenal lesions detected on CT or MRI. *J Nucl Med* 2001;42:1795–9.

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