CASE REPORTS

Imaging Of An Adrenal Cortical Carcinoma and Its Skeletal Metastasis

Walter E. Drane, Michael M. Graham, and Wil B. Nelp

University of Washington School of Medicine, Seattle, Washington

Though the typical scintigraphic appearance in adrenal cortical carcinoma is bilateral nonvisualization of the adrenal glands, we report a case with simultaneous visualization of both an adrenal cortical carcinoma and its skeletal metastasis using 6-β-[^{131}I]iodomethyl-19-norcholesterol.

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Nuclear adrenal imaging has proven to be highly accurate in distinguishing the various causes of Cushing's syndrome (*1,2*). Bilateral adrenal hyperplasia from excessive secretion of adrenocorticotrophic hormone (ACTH), adrenal adenoma, and adrenal cortical carcinoma each usually shows a distinct pattern. The typical pattern with adrenal cortical carcinoma is bilateral nonvisualization of the adrenal glands (*2,3*), but functional metastases have been visualized following the surgical removal of the primary tumor (*4–7*). We report a case with simultaneous visualization of both a primary adrenal cortical carcinoma and its skeletal metastasis using 6-β-[I-131]iodomethyl-19-norcholesterol, and discuss the significance of this finding.

CASE REPORT

A 66-yr-old woman was first referred for adrenal imaging in July, 1977. Since 1966 she had had progressive gain in weight. In 1971 she became hypertensive, and then developed diabetes mellitus, requiring daily insulin therapy. In September, 1977, an evaluation for Cushing's syndrome revealed truncal obesity with a "buffalo hump" deformity and moon face. Her serum cortisol level was elevated, as was her 24-hr urinary 17-hydroxy-corticosteroid (17-OH-CS) output, both before and after dexamethasone administration. Four days following the intravenous administration of 2 mCi of 6-β-[^{131}I]iodomethyl-19-norcholesterol (without dexamethasone suppression), a posterior abdominal image showed accumulation of radiotracer by a large left suprarenal mass, interpreted as a functioning left adrenal tumor (Fig. 1). The significance of the small focus of midline activity was not appreciated, and this activity, as well as the activity lateral to the right kidney, were felt to be within bowel.

A left adrenalectomy was performed, with removal of a 5 × 4.5 × 2.7 cm adrenal tumor weighing 32 grams. The tumor was adherent to the superior pole of the left kidney, and there was questionable invasion of the tumor capsule. Excluding its size and the suggestion of local invasion, the tumor grossly and histologically had the features of a benign adenoma. After surgery the patient became normotensive and no longer required insulin therapy. Serum cortisol and urinary 17-OH-CS returned to normal.

In July, 1980, diabetes mellitus and hypertension recurred. Serum cortisol was again elevated. Another adrenal scan was performed, which again demonstrated a small focus of radiotracer uptake in the midline (Fig. 2). In retrospect, this activity was identified on the previous adrenal scan, and the finding was interpreted as metastatic disease. A radionuclide bone scan was performed for correlation, and showed increased uptake in L1, probably the spinous process (Fig. 3). Radiographs of the lumbar spine confirmed the presence of an expansile lesion (Fig. 4).

A total L1 laminectomy was performed, and pathologic examination of the spinous process demonstrated adrenal cortical tissue histologically similar to that in the previous tumor. A postoperative TCT scan showed her normal right adrenal gland. The patient became normotensive and serum cortisol returned to normal, but diabetes mellitus persisted.

Over the course of the next year, the patient again exhibited signs and symptoms of Cushing's syndrome. Serum cortisol again became elevated. A follow-up adrenal image in June, 1981, showed a metastatic focus in the left pelvis (Fig. 5). This focus was felt to be in soft tissue, just anterior to the sacrum. A repeat radionuclide bone scan and a pelvic radiograph were normal. The right adrenal gland was still not visualized. Because of the patient's reluctance to undergo further surgery, she has been treated subsequently with antihypertensives and insulin, and is still alive at the time of this report.

DISCUSSION

Adrenal cortical carcinoma is a relatively rare malignancy, occurring in about two people per million (*8*). Analysis of the clinical and laboratory presentations of this tumor have shown that between 50 and 95% are functional, commonly resulting in

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For reprints contact: Wil B. Nelp, MD, Division of Nuclear Medicine, RC-70, University Hospital, University of Washington, Seattle, WA 98195.

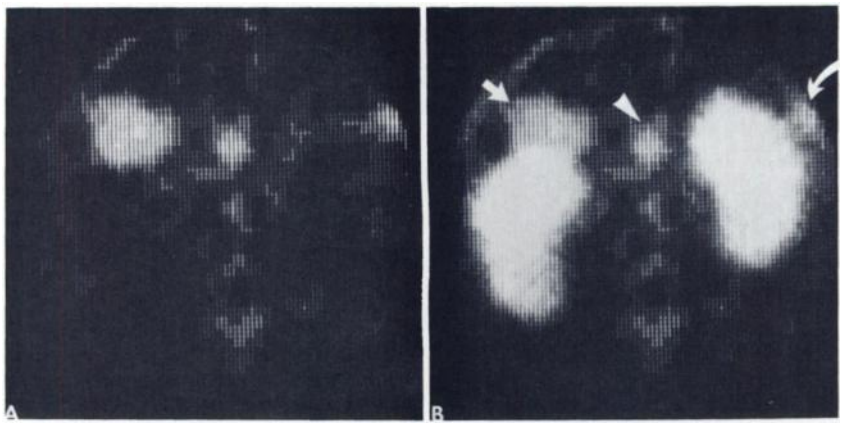

FIG. 1. Computer-acquired, posterior abdominal scintigram with 6-beta-[I-131]iodomethyl-19-norcholesterol scan (A). Computer-acquired, composite image formed with addition of DMSA renal scan (Tc-99m dimercaptosuccinic acid) to image (B). Large, left suprarenal mass is seen (arrow). Activity lateral to right kidney (curved arrow), and small midline focus (arrowhead), were felt to be within bowel (gallbladder had been removed). Right adrenal gland is not seen.

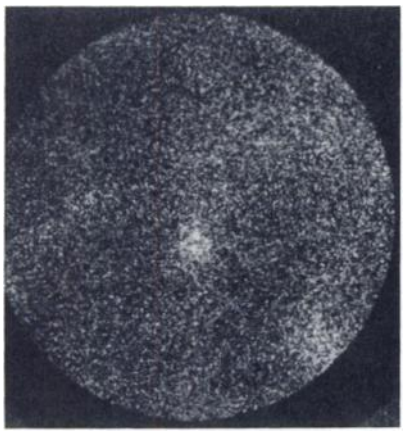

FIG. 2. Posterior abdominal analog image with 6-β-[^{131}I]iodomethyl-19-norcholesterol again shows small focus of increased activity in midline. Right adrenal gland is still not visualized. Radiotracer activity faintly outlines colon, normal finding.

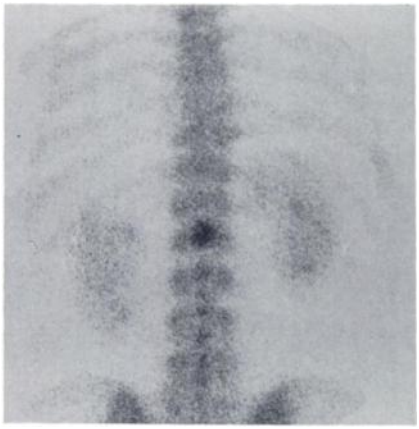

FIG. 3. Posterior bone scintigram shows increased uptake by L1 vertebra.

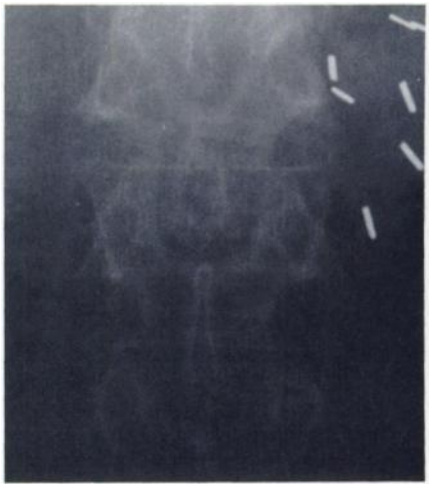

FIG. 4. AP radiograph of lumbar spine shows expansile mass in spinous process of L1.

Cushing's syndrome and/or virilization (*9–12*). Even though many of these tumors produce excessive cortisol, the typical finding with nuclear adrenal imaging is nonvisualization of the tumor with bilateral suppression (nonvisualization) of the normal adrenal tissue (*2,3*).

Schteingart and associates (*13*) determined the concentration of [I-131]iodocholesterol in neoplastic adrenal tissue and in uninvolved adrenal cortex obtained at surgery or autopsy within 2 to 3 wk after completion of adrenal imaging. They showed that the relative concentration of radiotracer was much lower in adrenal carcinoma than in either normal adrenal cortex or cortisol-secreting adrenal adenoma, despite the fact that their patients with carcinoma had evidence of increased steroid production. None of these carcinomas were visualized by scintigram. Presumably in these carcinomas, cortisol is derived primarily from endogenously produced cholesterol, with very little serum cholesterol entering the metabolic pathway.

The histological appearance of adrenal cortical carcinoma ranges from anaplastic to well-differentiated, and can be difficult to distinguish from benign adenoma. The most suggestive sign of

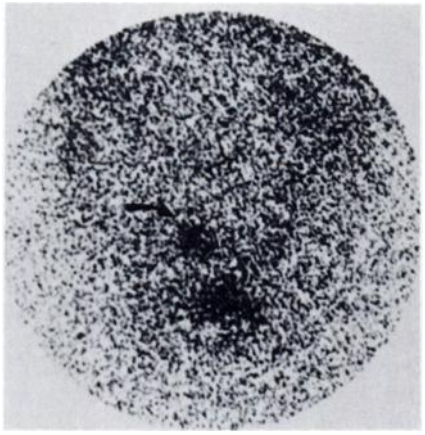

FIG. 5. Posterior 6-beta-[I-131]iodomethyl-19-norcholesterol scan identifies focus of increased uptake in left pelvis (arrow). Normal bladder activity is also seen. As occurred before, right adrenal gland was not seen.

malignancy is invasion of the capsule of the neoplasm (*14*), as was apparently present in our case. Likewise, the degree of cortisol production by adrenal cortical carcinoma is a spectrum. Unlike the typical findings with this disease, the malignancy in our patient was visualized, most likely because of its well-differentiated histology, with cells that retained the ability to trap exogenous, radiolabeled serum cholesterol, as in the metabolic pathway of normal adrenocortical tissue.

On occasion, metastatic adrenal cortical carcinoma has been imaged with [I-131]iodocholesterol (or its derivatives) after the removal of the primary tumor (*4–7*). Our case shows that simultaneous detection of both the primary tumor and metastatic sites is possible, and if there is suspicion of carcinoma, images of the chest, abdomen, and pelvis should be obtained. In addition, since adrenal cortical carcinomas typically achieve much larger sizes than adrenal adenomas before diagnosis, carcinoma should always be considered when nuclear adrenal imaging yields unilateral visualization of a markedly enlarged adrenal tumor.

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