Radionuclide Captopril Renography in Postpartum Renal Artery Aneurysms

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We report the case of a young woman with three left renal artery aneurysms, diagnosed immediately postpartum, associated with a de novo high blood pressure. To assess anatomical and functional characteristics of renal artery aneurysms, renal angiogram, MRI, intravenous pyelography, ultrasonography and radionuclide renography were performed. Two patent saccular renal artery aneurysms were demonstrated in the left kidney by renal angiogram. A larger, thrombosed aneurysm was also depicted on the left side on ultrasonography, MRI and renal angiogram. The larger aneurysm was responsible for renovascular disease of the middle third of the kidney, as demonstrated by captopril and baseline radionuclide renographic studies. It also impeded drainage of the lower pyelocalyceal group, without obstructing it, as shown by concomitant furosemide (Lasix) evaluations.

Key Words: renal artery aneurysms; renovascular hypertension; captopril radionuclide renography; pregnancy-related complications

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Renovascular disease is an important cause of arterial hypertension. Renal artery stenosis is usually caused by fibrodysplasia in young women or atherosclerosis in older patients. Saccular renal artery aneurysms are occasionally described, but are seldom seen in young patients. Rupture of renal or other visceral artery aneurysms is a catastrophic complication of pregnancy. We describe a hypertensive patient with three saccular renal artery aneurysms who survived pregnancy and delivery. The functional repercussion of the aneurysms was assessed by captopril/furosemide radionuclide renography.

CASE REPORT

A 24-yr-old woman had an unremarkable delivery at 40 wk of gestation. Pregnancy was uncomplicated, except for transitory elevation of blood pressure and nonspecific left flank pain at 36 wk. Immediately postpartum, arterial hypertension reappeared and persisted, blood pressure reaching 170/120 mmHg. Treatment was initiated with a calcium antagonist but had little effect on blood pressure. Three months postpartum, the patient was referred to our institution for further evaluation of hypertension. On admission, blood pressure was 130/100 mmHg. Physical examination was otherwise unremarkable. Serum creatinine and potassium levels were 91 μmole/liter (normal: 60–105) and 3.8 mmole/liter (normal: 3.5–5.2), respectively. Urine analysis was normal. Renovascular hypertension was suspected and captopril and baseline radionuclide renographic studies were performed. After intravenous injection of 370 Mbq (10 mCi) ^{99m}Tc-DTPA, a renogram was obtained: a 60 sec-flow study, followed by an analog study consisting of 9 frames of 60 sec each, 9 frames of 120 sec each and 9 frames of 120 sec each after intravenous injection of furosemide (40 mg Lasix). Digital data were then recorded and the differential ^{99m}Tc-DTPA function was derived from the background-corrected counts obtained between 1 and 3 min postinjection (*1*). Lasix was used in both studies to assure reproducibility, since the glomerular filtration rate (GFR) was to be measured. Lasix injection also helped identify and separate the different thirds of the left kidney in both studies.

First, the ACE-inhibitor radionuclide renography was performed 1 hr after administration of 25 mg Captopril. The initial differential uptake of ^{99m}Tc-DTPA was 49% on the left side and 51% on the right. A hypofunctioning zone was found in the left renal hilum and in the adjacent cortex of the middle third of the kidney; no significant abnormality was found in the upper third of the left kidney. Cortical accumulation of ^{99m}Tc-DTPA was demonstrated in the middle third of the kidney. Impaired drainage, without obstruction, was demonstrated in the lower third. No abnormality was noted on the right side (Fig. 1). One day later, a baseline radionuclide renogram was obtained and compared to the previous study. The initial differential uptake of ^{99m}Tc-DTPA was 52% on the left side and 48% on the right. No cortical abnormalities were observed in the middle third of the left kidney, but hypofunction was still demonstrated in the hilum (Fig. 2). As a result, renovascular disease was suspected in the middle third of the left kidney.

Ultrasonography with doppler examination revealed three aneurysms in the left kidney. The largest one, in the hilum, was not significantly patent.

The right renal angiogram was normal. The left selective renal angiogram demonstrated two saccular aneurysms that had developed on segmental arteries in the upper and lower third (Fig. 3A). A thrombosed segmental artery was found in the middle third of the kidney, near the hilum (Fig. 3A). MRI demonstrated the larger aneurysm, with minimal blood flow running through it (Fig. 3B).

Intravenous pyelography demonstrated contrast stasis in the lower pyelocalyceal group. Surgical repair of the three aneurysms was attempted but was unsuccessful. Left-side nephrectomy was performed. Blood pressure was normal thereafter, confirming the renovascular origin of the patient's hypertension. Pathological examination revealed vascular lesions consistent with neurofibromatosis. The patient, however, did not show any other sign or symptom of that disease.

DISCUSSION

The introduction of angiography has made the depiction renal artery aneurysms more frequent. Incidence varies from 0.3% to 2.5% in angiographic studies (*2–4*) and from 0.01% to 9.7% in autopsy studies (*2,4*). They are usually classified in four groups (*2,4–7*). Saccular aneurysms are the most frequent (70%–90%). Fusiform aneurysms (20%–25%) generally represent a poststenotic dilatation of the renal artery. Post-traumatic aneurysms and microaneurysms related to vasculitis are uncommon. Saccular aneurysms present as bulging lesions arising from the arterial wall, at renal artery bifurcations where congenital defects in the elastic tissue can be found. They are rarely larger than 2 cm (20%) or multiple (30%). With time, the aneurysms enlarge, due to increased mural tension and increased blood flow, and may rupture. Secondary atherosclerosis deposits and calcifications can occur (*4–7*). Fibromuscular disease and neurofibromatosis may cause such aneurysms (*6,8–12*). Sev-

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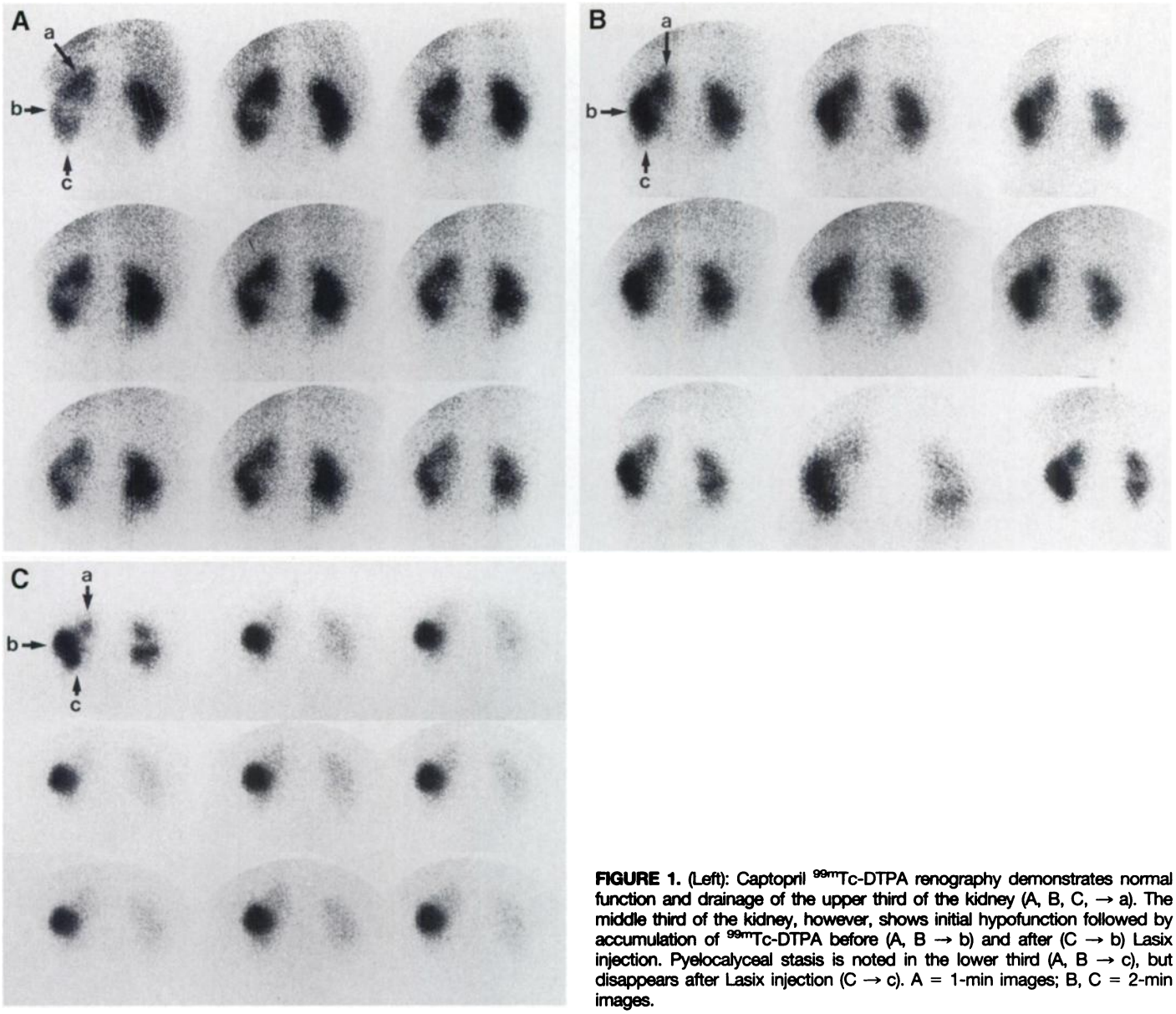

FIGURE 1. (Left): Captopril ^{99m}Tc-DTPA renography demonstrates normal function and drainage of the upper third of the kidney (A, B, C, → a). The middle third of the kidney, however, shows initial hypofunction followed by accumulation of ^{99m}Tc-DTPA before (A, B → b) and after (C → b) Lasix injection. Pyelocalyceal stasis is noted in the lower third (A, B → c), but disappears after Lasix injection (C → c). A = 1-min images; B, C = 2-min images.

enty to ninety percent of patients with saccular aneurysms develop hypertension (*4,6,8,9,13*). Surgical repair of these aneurysms or nephrectomy have been shown to improve or cure hypertension (*6,8,9,13,14–18*). Renovascular disease is thought to be secondary to compression of adjacent arteries (*14,15,19*), reduced renal flow distal to the aneurysm or thrombosis (*14,15,17,19*) or an unrecognized distal kink (*15*). Stanby et al. reported one case of saccular aneurysm detected by captopril renography (*12*), with no evidence of renal artery stenosis. In our case, as demonstrated by the radionuclide renographic studies, renovascular disease was only present in the middle third of the left kidney. The large aneurysm was almost completely thrombosed, causing severely reduced distal blood flow (MRI examination). Presence of collateral blood flow (renal angiogram) probably helped prevent infarction of this part of the kidney. In afferent renal artery stenosis, glomerular filtration rate (GFR) is maintained by Angiotensin II mediated constriction of efferent artery. Angiotensin-converting enzyme (ACE) inhibition prevents such compensatory mechanics and induces a fall in GFR (*20*). The initial hypofunction of the cortex of the middle third of the left kidney and subsequent ^{99m}Tc-DTPA accumulation was secondary to glomerular filtration rate reduction induced by captopril. ACE inhibition also induced reduction of the left differential function from 52% to 49%. The two remaining aneurysms were patent and did not produce renovascular disease.

Flank pain, abdominal pain or hematuria are found in less than 20% of the cases (*2–4,6,8,9,19*). Significant hydronephrosis is unusual (*21*). Our case demonstrated abnormal pyelocalyceal drainage of the lower pole but without obstruction. Rupture occurs in 5% of all cases but can be found in 25% of uncalcified aneurysms (*2,4,7*). In large, unselected, autopsy studies, however, rupture is rare (*3,22*).

Pregnancy warrants special consideration. In women under the age of 40, visceral artery aneurysms are often discovered when they rupture. Almost 50% rupture during pregnancy (*23*). Twenty-seven cases of renal artery aneurysms related to pregnancy have been described in the literature (*7,12,24–28*). Twenty-five ruptured—1 during the second trimester, 22 during the third trimester and 2 postpartum. Only two aneurysms were discovered intact postpartum (*12,27*). The vast majority of patients do not have any symptoms until rupture. Hypertension is absent, except in our and the case of Stanby et al. (*12*). Occasionally, vague flank pain or hematuria are reported. Rupture is a catastrophic event characterized by acute, severe flank pain, shock and a high mortality rate, both for the mother

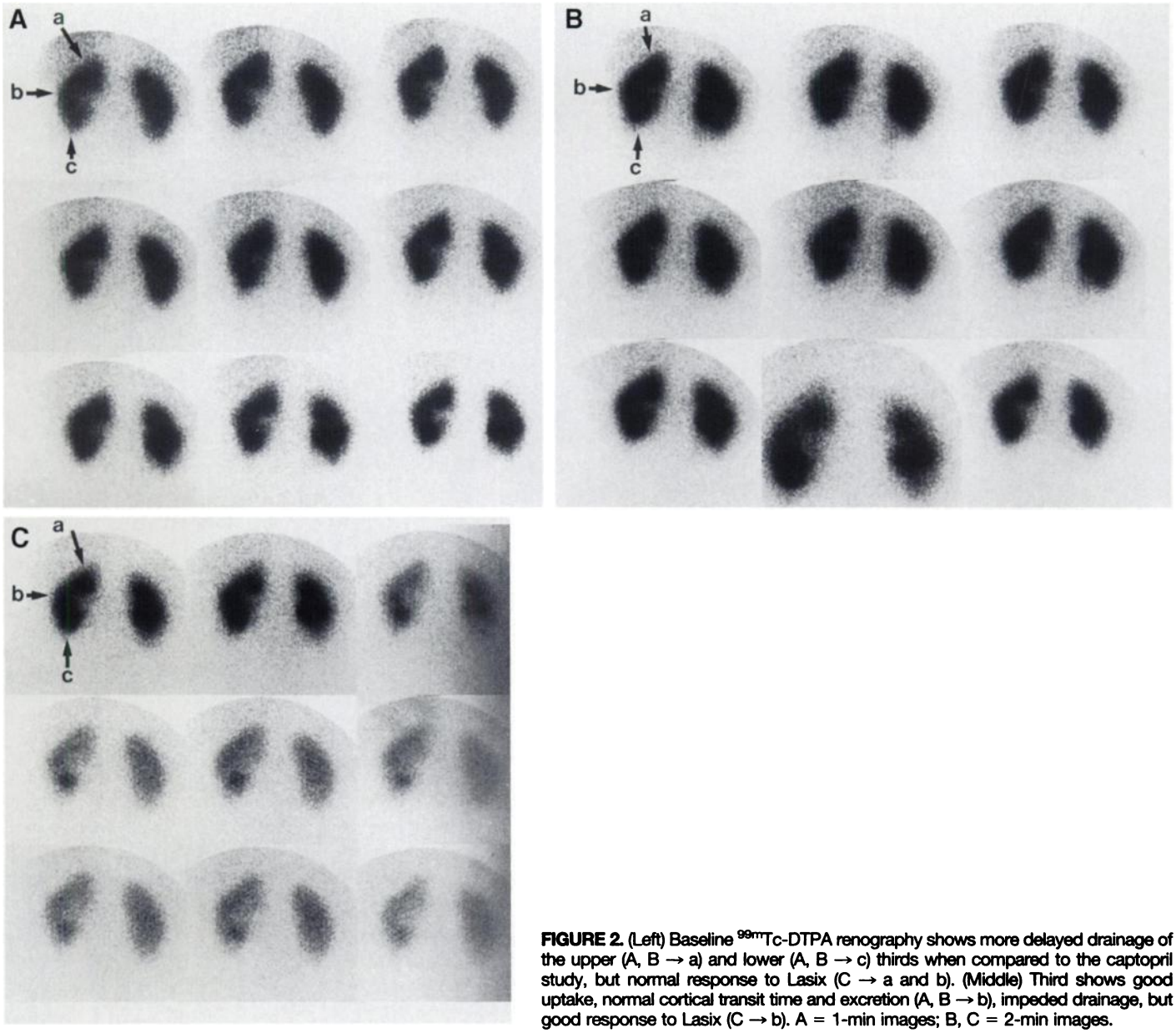

FIGURE 2. (Left) Baseline ^{99m}Tc-DTPA renography shows more delayed drainage of the upper (A, B → a) and lower (A, B → c) thirds when compared to the captopril study, but normal response to Lasix (C → a and b). (Middle) Third shows good uptake, normal cortical transit time and excretion (A, B → b), impeded drainage, but good response to Lasix (C → b). A = 1-min images; B, C = 2-min images.

and for the baby. Fortunately, earlier recognition of this complication has allowed swifter intervention, consisting of either emergency nephrectomy or primary repair, and has permitted better survival since 1970. The two "intact" aneurysms discovered postpartum in our patient were repaired uneventfully. The majority of aneurysms are on the left side (*7*). Pathological examination (*24*) usually does not demonstrate any particular underlying pathology. Fibrodysplasia or neurofibromatosis are occasionally found (*12,24*). Pregnancy-related factors that increase the risk of rupture are numerous and

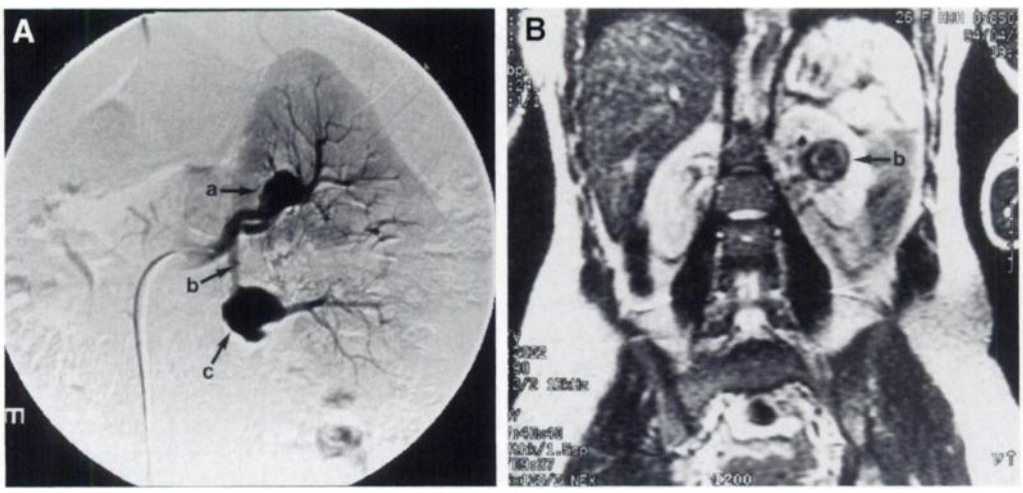

FIGURE 3. (A) Two saccular aneurysms (2- and 2.5-cm, respectively) are demonstrated angiographically in the left kidney (a, c). Thrombosis of a segmental artery with collateral circulation and mass effect is found in the middle third (b). (B) Coronal T2-weighted MR image of the left kidney demonstrates a 4-cm partially thrombosed calcified saccular aneurysm (b).

include hormonally mediated vascular relaxation, intimal hyperplasia and fragmentation of reticulum fibers (progesterone increases noncollagenous protein deposits and estrogen decreases elastin-collagenous content). Also, we can observe increased blood volume and increased cardiac output, and vascular strain due to compression of the inferior vena cava and aorta by the gravid uterus, especially in late pregnancy (*7,23,26,28*). Therefore, removal of renal artery aneurysms in women of child-bearing age is largely recommended, especially if pregnancy is considered. Surgery is also preferred in patients with aneurysms larger than 1.5 cm, uncalcified aneurysms, lesions increasing in size with time and in symptomatic or uninephric patients (*2,6,7,19*).

CONCLUSION

Captopril renal scintigraphy with differential assessment of various portions of an ischemic kidney is useful to evaluate the significance of segmental renal artery disease. In this particular case, a renal artery aneurysm was responsible for renovascular disease in one-third of the left kidney.

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Bone Scintigraphy in Hungry Bone Syndrome Following Parathyroidectomy

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A 59-yr-old man with chronic renal failure was admitted for evaluation of generalized skeletal pain and frontal bone mass, which was lytic on radiography. Bone scintigraphy demonstrated several foci of moderately increased uptake, without involvement of the skull mass. Radiographs of these lesions were compatible with brown tumors. Serum parathormone level was elevated and CT demonstrated a lower right cervical mass, consistent with parathyroid tumor. Following the removal of the mass and decrease in parathormone levels, the patient suffered from a prolonged period of hypocalcemia and his bone pain worsened. Repeat bone scintigraphy showed an increase in the number and intensity of the areas of focal uptake, consistent with hungry bone syndrome. This flare-up phenomenon is due to an increase in bone metabolism and is an uncommon finding following parathyroidectomy for primary hyperparathyroidism.

Key Words: hyperparathyroidism; brown tumor; bone scintigraphy; parathyroidectomy; hungry bone syndrome

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CASE REPORT

A 59-yr-old man in moderate chronic renal failure of unknown etiology complained of generalized bone skeletal pain and recent development of a mass in his forehead. On physical examination, a solid mass with bone consistency, as well as a large neck mass, were palpated. Radiography revealed a large lytic lesion in the left frontal bone (Fig. 1). Bone scintigraphy with ^{99m}Tc-MDP demonstrated diffuse tracer activity in the skull with questionable uptake above the left orbit. Several foci of faint to moderately increased uptake in the sternum, ribs, left shoulder and right sacroiliac joint were visualized (Fig. 2), and were lytic on radiography. Relevant laboratory tests included hemoglobin 10.1 g/liter, WBC 8540/mm^3, blood urea nitrogen 40 mg/dl, serum creatinine 3.5 mg/dl, creatinine clearance