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# Bilateral Adrenal Pheochromocytoma Diagnosed by Somatostatin Receptor Scintigraphy

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**Abstract** **Case Report**

Pheochromocytoma is a rare and severe pathology; producing an excess of catecholamines. Adrenal involvement is most often unilateral. In 10% of cases, bilateral localization is observed, most often in the context of a familial disease. However, sporadic bilateral forms can be observed. The interest of our work is to highlight the particularities of this pheochromocytoma, and the contribution of metabolic imaging in the assessment of its localization. In this regard, we report the observation of a 30-year-old patient who was hospitalized for exploration of secondary arterial hypertension, in a context of elevation of methoxylated derivatives to 46 times normal. Abdominal CT noted the presence of two bilateral adrenal masses. The NEM 2 assessment is negative. Somatostatin receptor scintigraphy (SRS) showed hyperfixation at the level of the two bilateral adrenal masses. The diagnosis of bilateral pheochromocytoma was made. Bilateral adrenalectomy was performed. The diagnosis of bilateral pheochromocytoma requires confirmation of the secretory nature of both adrenal tumors. Scintigraphy can objectify the neuroendocrine nature of both adrenal masses. The search for an underlying genetic disorder must be systematic.

**Keywords:** Pheochromocytoma, Adrenal, Scintigraphy.



## INTRODUCTION

Pheochromocytoma is a rare neuroendocrine tumor originating from chromaffin cells, most commonly located in the adrenal medulla, and is marked by excessive catecholamine production. Bilateral pheochromocytomas occur in approximately 10% of cases and are frequently associated with hereditary syndromes. The presence of bilateral tumors should prompt investigation for an underlying germline mutation, which can be identified in up to 80% of cases. Bilaterality introduces specific challenges regarding diagnosis, assessment of malignancy potential, and genetic evaluation particularly in patients without a known family history. Recognition of familial forms requires careful evaluation of the patient's family history and a thorough clinical workup [1, 2]. Diagnostic confirmation is based on the measurement of urinary or plasma catecholamines, their precursors, or metabolites. Advances in imaging techniques, particularly magnetic resonance imaging (MRI) and metaiodobenzylguanidine (MIBG) scintigraphy, have significantly improved tumor localization.

Therapeutic management requires a multidisciplinary approach. Histopathological examination of the surgical specimen provides definitive confirmation of the diagnosis [3].

The interest of our work is to highlight the particularities of this pheochromocytoma, and the contribution of functional imaging in the assessment of its location.

## CASE REPORT

We report the case of a 30-year-old man from a non-consanguineous marriage with a medical history of type 1 diabetes mellitus under insulin, hypertension under dual therapy, and a hemorrhagic stroke. He had no family history of neuroendocrine tumors or autoimmune diseases. The patient presented to the hospital for investigation of secondary hypertension.

Physical examination revealed a rapid and pounding heart rate, excessive sweating, and headache with abdominal pain on palpation.

Laboratory results demonstrated an elevation of methoxylated derivatives to 46 times the normal range.

– A CT Scan Revealed:

Large adrenal masses measuring 3 x 7 x 7.2 cm on the right and 4.2 x 4.5 x 7.5 cm on the left, well demarcated, spontaneously hypodense, heterogeneous enhancement, central necrosis, and small calcification on the left.

– Given this bilateral involvement, somatostatin receptor scintigraphy was performed, with planar acquisition 1 hour and 4 hours after IV injection of 17 mCi of 99mTc-labeled Tektrotyd; and with SPECT/CT acquisition of the abdominopelvic region:

The 1-hour and 4-hour full-body scans revealed a small focus of hyperuptake in the posterior epigastric projection. Moderate bilateral uptake enhancement was observed in the suprarenal regions **(Figures 1,2)**.

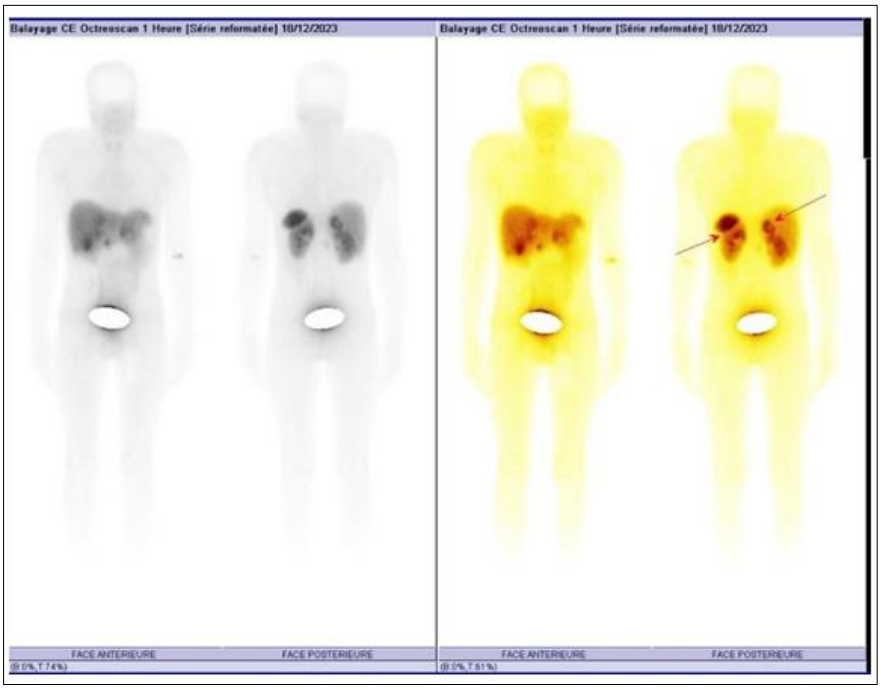


**Figure 1: Full body scan 1 hour**

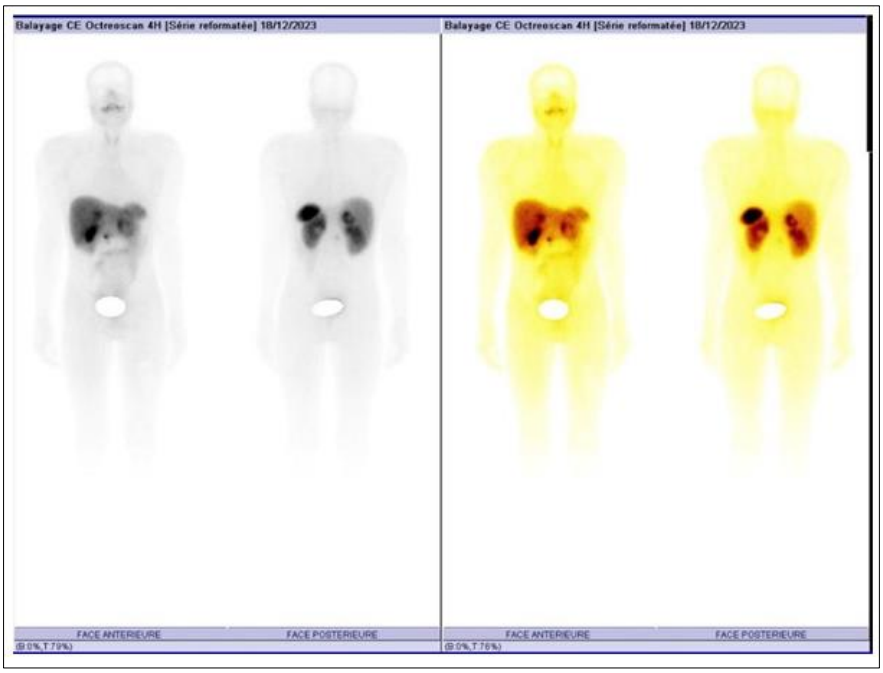


**Figure 2: Full body scan 4 hour**

Computed tomography (SPECT/CT) of the abdominopelvic region **(Figures 3,4)**: Suprarenal hyperfixations on planar images correspond to adrenal masses on T2MP-CT fusion images, presenting central hypofixation (central necrotic areas), consistent in the

context with pheochromocytomas. No secondary localization.

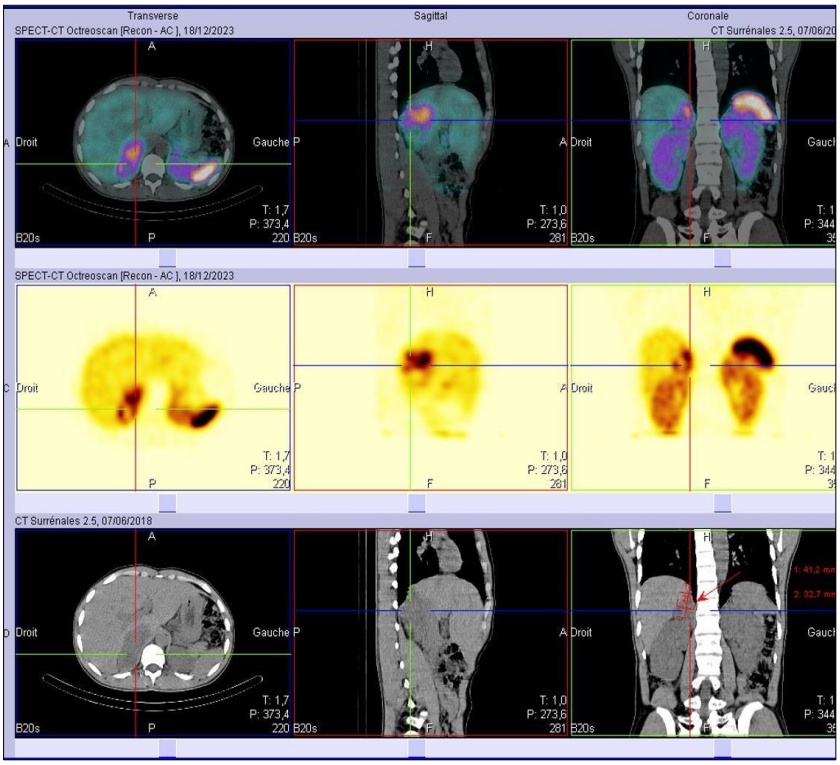


**Figure 3: CT-combined tomoscintigraphic sections (SPECT/CT) of the abdominopelvic region, showing the pheochromytoma at the level of the right adrenal gland**

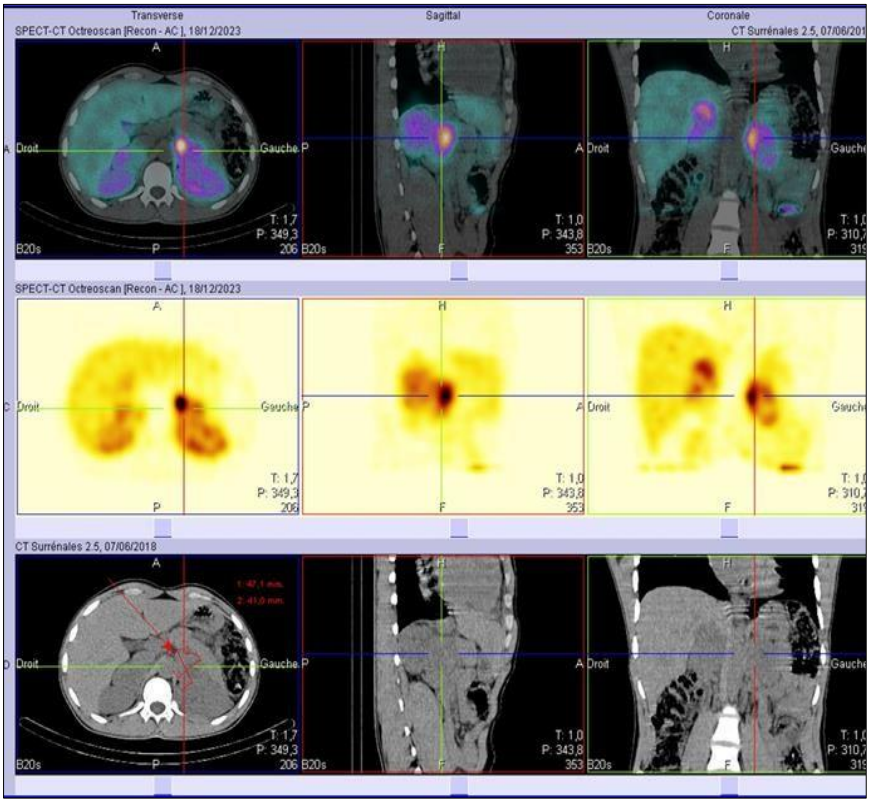


**Figure 4: CT-combined tomoscintigraphic sections (SPECT/CT) of the abdominopelvic region, showing the pheochromytoma at the level of the left adrenal gland**

The diagnosis was made: bilateral pheochromocytoma in its sporadic form, given the absence of any particular family history. Treatment was bilateral adrenalectomy in two stages by laparoscopic approach after blood pressure equilibration. The surgical specimens were sent to pathology for histological study, which confirmed the diagnosis. The postoperative course was uneventful. The outcome was favorable under replacement therapy. No recurrence was observed.

## DISCUSSION

Pheochromocytomas and abdominal paragangliomas are catecholamine-producing tumors that arise from paraganglia cells derived from the neural crest. Paraganglia of the sympathoadrenal neuroendocrine system are distributed along the para-vertebral and para-aortic axis, from the base of the skull to the pelvic floor **(Figure 5)**. The annual incidence of pheochromocytoma is estimated to be 1–2/million population. The traditional rule of thumb is that approximately 10% of pheochromocytomas are malignant, 10% are bilateral, 10% are extra-adrenal (of which 10% are extra abdominal), 10% are not associated with hypertension, and 10% are hereditary [4]. A careful family history and a thorough physical examination are necessary to recognize a hereditary disease. However, family history may be overlooked or missing in cases of primary mutation [5].

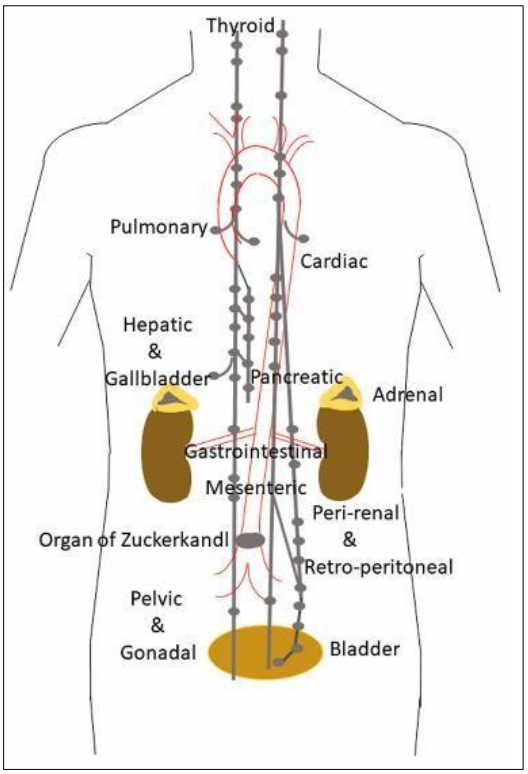


**Figure 5: Distribution of paraganglia tissue in the body and location of pheochromocytomas and paragangliomas**

23I -metaiodobenzylguanidine (123I-MIBG) scintigraphy is commonly used for functional imaging of pheochromocytomas and paragangliomas (PPGLs), tumors arising from respective adrenomedullary and extra-adrenal chromaffin cells. It has the advantage of performing whole-body imaging, which is very useful for detecting ectopic tumors, searching for metastases or recurrences of malignant pheochromocytoma. Combined with CT scanning, it increases the sensitivity of topographical investigation from 90% to nearly 100%. Its sensitivity is superior to that of MRI for extra-adrenal tumors (88% vs. 75%), as is its specificity (100% vs. 80%) [6-8]. [123I] MIBG uptake was significantly correlated with tumor size and epinephrine production and was significantly greater in benign, sporadic, intraadrenal, and unilateral tumors [9].

Overall, the scintigraphic modality with the highest sensitivity for localizing PHEO was [18F] DA PET (75.4%), followed by [123I]-MIBG (63.4%) and SRS (64%) [10].

Comparing in a few reports the diagnostic accuracy of SRS and [123I]- or [131I]-MIBG scintigraphy in patients with metastatic pheochromocytoma, SRS was found to have an overall higher detection rate: SRS detected up to 87% of lesions, while [123I]-MIBG localized only 57% [11,12].

The study by Ioannis Ilias *et al*., showed that in the diagnostic evaluation of pheochromocytoma, [18F]-DA PET and [123I]-MIBG are more sensitive than SRS for detecting non-metastatic primary adrenal pheochromocytoma. For metastatic pheochromocytoma,

[18F]-DA is more sensitive than SRS, and both are superior to [123I] MIBG [10].

The diagram in **Figure 6** summarizes the diagnostic algorithm for pheochromocytoma [13].

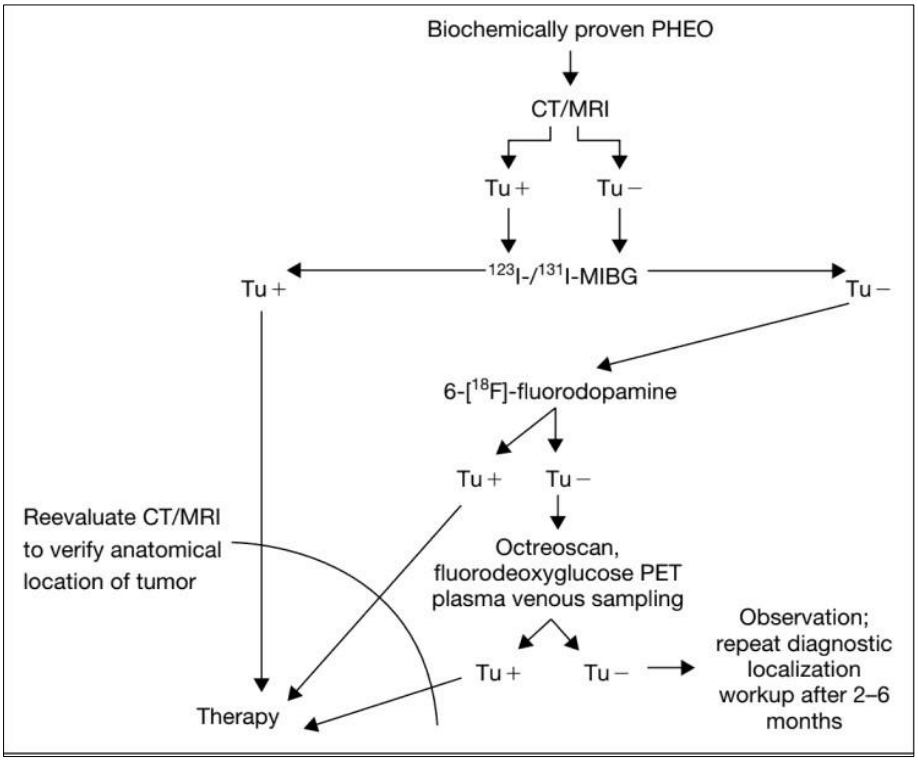


**Figure 6: Proposed diagnostic algorithm for pheochromocytoma**

In conclusion, pheochromocytoma is a rare and potentially severe condition. Early onset, bilaterality, and association with high-risk clinical features contribute to its overall severity. This case highlights the importance of effective multidisciplinary collaboration in the management of complex presentations such as bilateral pheochromocytoma, ultimately leading to a successful therapeutic outcome. MIBG scintigraphy or somatostatin receptor imaging plays a key role in confirming the neuroendocrine nature of bilateral adrenal masses, thereby helping to exclude other types of adrenal incidentalomas. A systematic search for an underlying genetic disorder is essential in all such cases.

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**Authorship Contributions:**
Medical Practices**:** H.A., S.E.A., M.A.B., A.M., Concept: H.A., Design: H.A., Data Collection or Processing: H.A., Analysis or Interpretation: H.A., S.E.A., M.A.B., A.M., Literature Search: H.A., Writing: H.A.

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**Declarations**

**Conflict of Interest**:

All the authors (Hamza Alaoui, Saad Eddine Abaid, Mohammed Aziz Bsiss, Aboubaker Matrane) declare that there is no confict of interest that could be perceived as prejudicing the impartiality of the research reported.

**Informed Consent**: Written informed consent was obtained prospectively from the patient to use their clinical data.

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