

## Oncology

## Giant bilateral adrenal myelolipoma: Case presentation and a brief literature review

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## Introduction

First described in 1905, adrenal myelolipomas are rare, hormonally inactive, benign tumors that are composed of mature fat tissue and hematopoietic elements. Most adrenal myelolipomas are small (4 cm) and asymptomatic (70%). The tumor is most frequently diagnosed in patients between the fifth and seventh decade of life.<sup>1</sup> We present a case of a giant bilateral adrenal myelolipoma found in a patient who initially present with abdominal pain.

## Case report

A 52-year-old man, hypertensive, non-insulin dependent diabetic, with left hemiplegia due to a previous stroke 4 years earlier, was referred to the urology clinic due to a complaint of diffuse insidious, sporadic, self-limited abdominal pain associated with recurrent vomiting since February 2017. There was no other associated symptoms. On physical examination, he was in good general condition and without palpable abdominal masses. Abdominal and pelvic CT showed retroperitoneal expansible lesion in the adrenal glands with a predominance of fatty tissue, measuring 13.3 × 9.3 cm in the left adrenal and 5.2 × 5.0 cm in the right, suggestive of bilateral adrenal myelolipoma (Fig. 1). No abnormalities in laboratory tests, as well as in hormonal investigation for adrenal masses were found (non-secretory neoplasia).

The Patient was submitted to open left adrenalectomy by subcostal incision in May 2017, with surgical time of three hours. The procedure was performed without complications (Fig. 2). The patient remained in the intensive care unit bed until the second postoperative day. The

hospital discharged was on the fourth day after surgery.

After histopathological analysis, the diagnosis of adrenal myelolipoma was confirmed, the piece measuring 16 × 13 × 7 cm and weighing 712g (Fig. 3).

The clinical follow-up of the myelolipoma in right adrenal was chosen due to the resolution of the abdominal pain. On CT scan, in October 2017, there was slight growth in the right lesion, measuring 5.8 × 5.5 cm. Patient remains in follow-up.

## Discussion

The incidence of myelolipoma increases with age. Some publications report that myelolipoma is three times more common on the right side than on the left.<sup>2</sup> About 12% cases are bilateral.<sup>2</sup> Myelolipomas are relatively slowly growing tumors and tumors exceeding 10 cm in diameter are called giant myelolipomas.<sup>3,4</sup> Big tumors can become symptomatic. Symptoms may include nonspecific abdominal pain, constipation, vomiting, hematuria, or renovascular hypertension because of intratumoral hemorrhage or compression of peritumoral tissue.<sup>3,4</sup>

Though several hypothesis have been proposed, the etiology of myelolipoma remains obscure. It may originate from remnants of fetal bone marrow, embolism of bone marrow cells, and hyperplasia of heterotopic reticulum cells.<sup>3,4</sup> Another major group of hypotheses relates to hormonal pathways especially bilateral myelolipomas. They have been associated with overproduction of adrenocorticotrophic hormone as in Cushing disease and same cases of Congenital adrenal hyperplasia.<sup>4,5</sup> It might be possible that both altered mesenchymal stem cell functioning and hormonal stimuli act together in the pathogenesis

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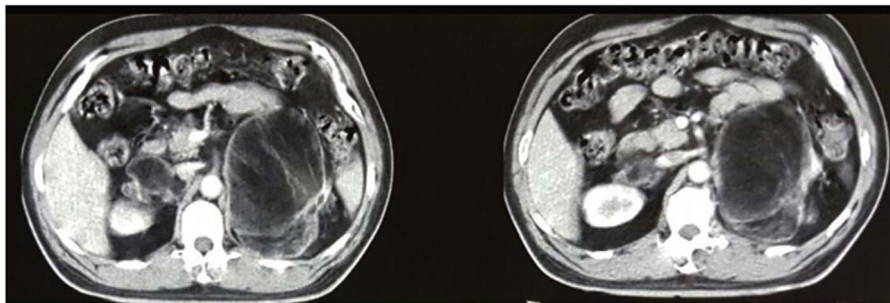


Fig. 1. CT: Bilateral adrenal myelolipoma.



Fig. 2. Giant left adrenal myelolipoma, surgical specimen. Macroscopic aspect of giant adrenal myelolipoma. This tumor is yellowish and soft, well delimited, surrounded by a capsule, with areas of cystification and hemorrhage.

of myelolipoma.<sup>4</sup> Adrenal myelolipomas, are generally hormonally inactive, as in this case, although there are case reports of their association with overproduction of adrenal hormones.<sup>4,5</sup>

In the past, these tumors were accidentally discovered at autopsy. Today, they are found much more frequently and incidentally, mainly because of the widespread use of noninvasive imaging with ultrasonography, computed tomography, and magnetic resonance imaging.<sup>5</sup> At computed tomography, a myelolipoma typically appears as a well-circumscribed mass that contains variable amounts of soft tissue and

macroscopic fat with negative Hounsfield Unit typically in a range - 40 or less.

Acute hemorrhage, although rare, is the most significant complication especially in large myelolipomas, and it can be manifested with pain in the back, epigastrium, or flanks, associated with nausea, vomiting, hypotension and anemia. Surgical resection is recommended in these cases.<sup>3</sup> Transarterial embolization with polyvinyl alcohol (PVA) particles or gelatin sponge particles could be used for successful hemostasis before adrenalectomy in patients with retroperitoneal hemorrhage from adrenal myelolipomas.

Due to the lack of formal guidelines regarding the management of adrenal myelolipomas, the decision should be made on a case-to-case basis. If surgical removal is indicated because of significant growth, symptoms (e.g., abdominal discomfort) or hormonal activity, laparotomy or laparoscopy could be performed.<sup>4</sup>

In the English literature, approximately 40 cases of bilateral myelolipomas were reported.<sup>1</sup> This is a particular case with a bilateral giant adrenal mass managed surgically in the left side without complications.

## Conclusion

Adrenal myelolipomas are rare, benign neoplasms of the adrenal gland with varied clinical presentations. This case presents an extremely rare diagnosis of bilateral myelolipoma with giant mass on the left side accompanied by abdominal pain symptoms. In this particular case, we also highlight the lack of hormonal dysfunction. The management of myelolipoma should be done on a case-to-case basis. The available literature is limited to case reports and short series from referral centers.

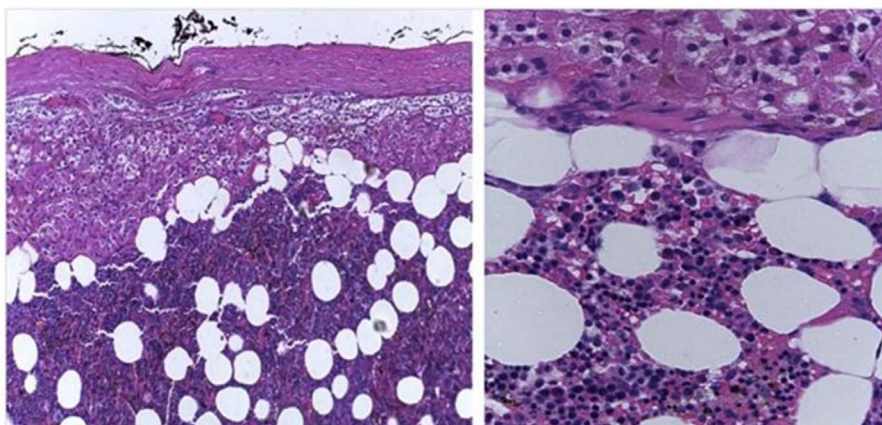


Fig. 3. Myelolipoma, histopathological confirmation. Histological sections of the tumor showing mature adipose tissue interspersed by extra-medullary hematopoietic tissue, compressing the adrenal glandular tissue and surrounded by fibrous capsule (Hematoxylin-Eosin, 200X and 400X).

**Interest conflicts**

No

**Appendix A. Supplementary data**

Supplementary data related to this article can be found at <http://dx.doi.org/10.1016/j.eucr.2018.03.008>.

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