CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 12 (2015) 31–36

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

The management of bilateral myelolipoma: Case report and review of the literature

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ARTICLE INFO

Article history:
Received 13 February 2015
Received in revised form 12 April 2015
Accepted 13 April 2015
Available online 7 May 2015

Keywords:
Myelolipoma
Adrenal gland
Adrenal tumor
Bilateral myelolipoma
Adrenal myelolipoma

ABSTRACT

INTRODUCTION: Bilateral adrenal myelolipoma is a rare benign neoplasm. We presented the case of a young man affected by a bilateral myelolipoma and the analysis of the literature of bilateral cases of myelolipoma. Our purpose is to give a suggestion of clear terms of reference regarding the management of this kind of bilateral neoplasm.
PRESENTATION OF CASE: We reported the case of a 41-year-old healthy man complained of abdominal pain in the upper quadrants. No significant alterations were found in routine blood and endocrinological tests. The imaging (CT and MRI) showed a huge right adrenal mass and a smaller lesion at the left adrenal gland. The preoperative pathological characterization was suggestive for a myelolipoma. A right open adrenalectomy was performed, and a radiological surveillance was planned for the left tumor. The pathological exam confirmed the diagnosis.
DISCUSSION: In literature, there are 36 cases described. The clinical presentation consisted of symptomatic tumors, incidentally diagnosed lesions or myelolipomas in patients with an associated endocrinal disorder. Symptomatic tumors or those bigger than 7 cm, because of the potential risk of rupture, are usually treated surgically. In smaller (<7 cm) and asymptomatic ones the surgical treatment is not univocal.
CONCLUSION: In the setting of the surgical treatment, it is important to preserve in some way the hormonal function. For that reason, the bilateral adrenalectomy has to be reserved for symptomatic or sizeable (>7 cm) cases. As far as we know, this is the first review on bilateral myelolipomas.

1. Introduction

The adrenal myelolipoma is a rare benign tumor of the adrenal gland. The incidence in old autopsy-series is about 0.08–0.25%; however, nowadays it is being diagnosed more and more frequently because of the widespread use of ultrasound, computed tomography (CT) and Magnetic Resonance Imaging (MRI). It represents about 3–5% of primary tumors of the adrenal gland [1].

In general these tumors are asymptomatic, unilateral and small (on average less than 4 cm). Sometimes they can cause symptoms because of bulk effects or rupture and also reach impressive dimensions (biggest tumor described measured 31 × 24.5 × 11.5 cm and weighed 5900 g) [2]. In some cases, they can be found bilaterally.

So far, the pathogenesis of the myelolipoma is unknown, but an adrenocortical metaplasia in response to stimuli like necrosis, inflammation, infection or stress [3] is the most common theory. Another hypothesis identifies a stressful lifestyle and a messy diet as risk factors [4].

Histologically, it is composed of variable mixture of mature adipose tissue and hematopoietic elements. We describe the case of a young patient with bilateral myelolipoma and present an analysis of literature regarding the reported cases of bilateral myelolipoma. The goal of our study was to attempt to identify clinical features and treatment criteria in this setting. To the best of our knowledge, this is the first review on bilateral cases.

2. Case report

A 41-year-old healthy man complained of abdominal pain after eating in the upper quadrants from 6 months, without other symptoms. No significant alterations were found in a routine blood test. Endocrinological tests and assay of urinary catecholamines did not show significant alterations.

A CT (Fig. 1) highlighted a huge right adrenal mass sized about 17 × 12 cm (-20HU) and a similar lesion of 2.3 × 2.5 cm (-5HU) in the left adrenal gland. MRI (Fig. 2) confirmed the presence of

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http://dx.doi.org/10.1016/j.ijscr.2015.04.021

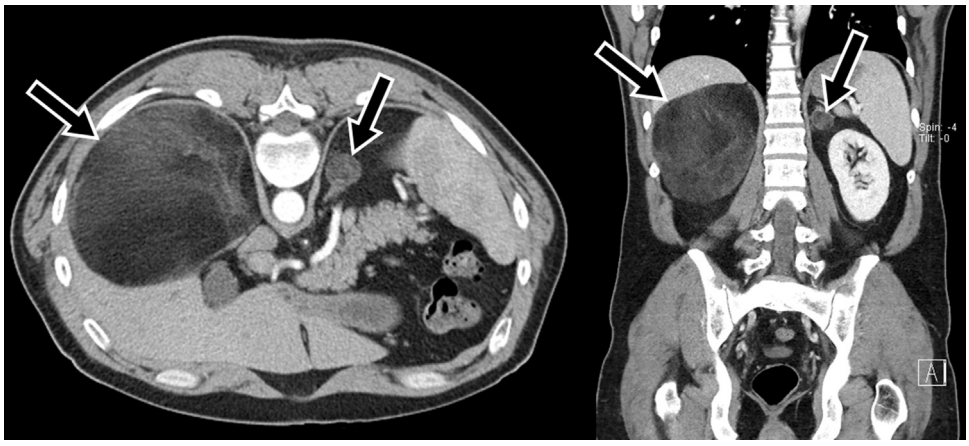

Fig. 1. CT scan.

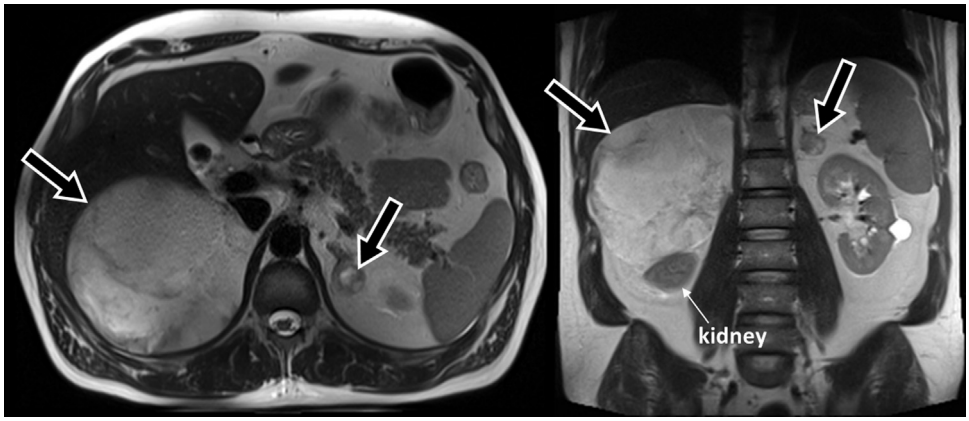

Fig. 2. MRI.

bilateral adrenal mass containing mainly fat with multiple septa, both consistent with myelolipomas.

A CT-guided biopsy of the major lesion showed the presence of fibroadipose tissue with focal hemosiderosis, without evidence of neoplastic cells or hematopoietic tissue.

In order to remove the huge symptomatic lesion, we performed a right open adrenalectomy (Fig. 3), using a Chevron incision. In order to safely and easily remove the main adrenal lesion (especially to achieve an adequate control of the adrenal vein), the Kocher maneuver have been required, together with the complete mobilization of the right liver and the right colic flexure, mainly because the mass being adherent to the right aspect and rear of the inferior vena cava (Fig. 4). Because of the asymptomatic feature and the small diameter, in order to preserve the adrenocortical function, we decided not to surgically remove the myelolipoma on the left side, monitoring over time its behavior by radiological surveillance.

The postoperative period was uneventful. After one-year follow-up with CT-scan, we did not find relapse on the right side, where the big myelolipoma was removed, and the left myelolipoma remained unchanged.

Macroscopically (Fig. 3), the specimen was a capsulated nodular lesion of 17 cm in diameter, weighing 1.170 kg. The color of the capsule was yellow mottled. To the cut, the tissue appeared fairly uniform, adipose and sallow, with hemorrhagic circumferential zones. The lesion seemed to be originated by the adrenal medulla with the capsule consisting of the cortex.

Microscopically (Fig. 5), the specimen showed a well-defined margins tumor, partly bounded by thin connective tissue capsule, largely consisting of mature adipose tissue with extensive hemosiderosis. Some areas of hematopoiesis have been identified, together with the presence of the three hematopoietic lineages. In peripheral areas have been discovered the presence of adrenal tissue with no significant histological changes.

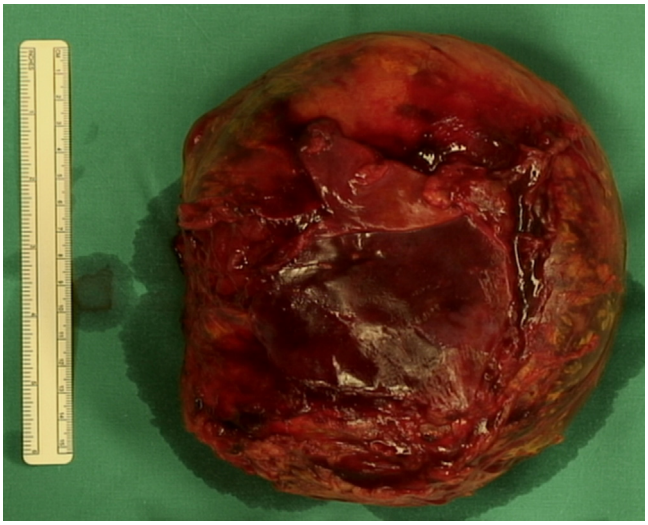

Fig. 3. Resected specimen.