

Case Report

Adrenal collision tumor: a case report of the coexistence of myelolipoma and ganglioneuroma

Qinglai Tang*, Junbiao Ji*, Xingzhu Zhou, Rongzhen Tao

Department of Urology, The Affiliated Jiangning Hospital with Nanjing Medical University, Nanjing, Jiangsu, China.

*Equal contributors.

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Abstract: Adrenal collision tumor (ACT), where distinct tumors can coexist and do not intermingle in the same adrenal gland, is rarely reported and its cause and prevalence are unclear. Few papers have reported a connection between myelolipoma and adrenal ganglioneuroma. Case Report: A 40-year-old man had a physical examination one month prior to surgery. Physical examination as well as the laboratory data involving endocrine studies showed normal findings. MRI (magnetic resonance imaging) revealed a mixed signal lesion in the right suprarenal fossa. A laparoscopic left adrenalectomy was conducted. Pathologic examination revealed two distinct tumors: myelolipoma and ganglioneuroma. Conclusion: The two kinds of tumors are rarely seen in the adrenal gland and they exceptionally exist as ACT only. This case should arouse our attention in future clinical work.

Keywords: Adrenal collision tumor, myelolipoma, ganglioneuroma, pathologic

Introduction

Adrenal collision tumor (ACT) is an infrequently described tumor entity, containing two different neoplasms that exist together with each other in a single adrenal mass [1, 2]. However, ACT's true prevalence is still unclear, despite the relatively great incidence of metastatic lesions and benign ones of adrenal glands. Adrenal myelolipoma, including mature adipose and hematopoietic tissues, is a benign neoplasm and hormonally inactive. Oberling first proposed the concept of myelolipoma in 1929 [3]. With the development of histopathology, we found that myelolipoma can coexist with a variety of other pathologic conditions of adrenal, e.g. adrenal cortical hyperplasia, enzyme deficiencies, and neoplasms of adrenal cortex. On the other hand, ganglioneuroma mainly originates from primordial neural crests which are composed of nerve fibers, mature Schwann cells, and a varying amount of ganglion cells [4]. It occurs mostly in mediastinal and aortocaval sympathetic ganglia but less frequently in the adrenal gland. In most cases, the two neoplasms exist as a single mass. This study reports a case of the coexistence of myelolipoma and ganglioneuro-

ma and reviews the literature to raise awareness of this extremely rare combination.

Case report

A 40-year-old Chinese man had a physical examination one month before surgery in the Affiliated Jiangning Hospital with Nanjing Medical University and had abnormal imaging findings. The imaging was found by MRI (magnetic resonance imaging, MRI) which revealed a mixed signal lesion in the right suprarenal fossa. The diameter of the mass was about 4 cm and had lesions with low and high signal intensities on T1WI and T2WI, respectively (**Figure 1**). Concomitant symptoms were not found, such as: epigastric pain, nausea, or weight loss. He denied a medical history of hypertension or diabetes. In addition, remarkable differences were not found from his review of systems and physical examination.

The results of laboratory tests showed the chemistry panel and complete blood cell counts were normal. Endocrine tests were also done, and the results did not show significant steroid hormone or catecholamine excess. The patient

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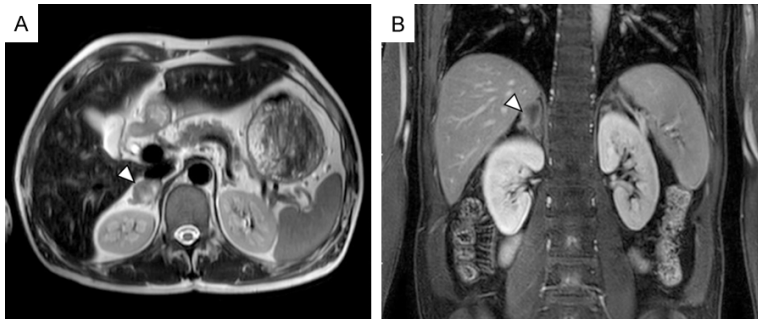


Figure 1. MRI revealed a mixed signal lesion in the right suprarenal fossa. The diameter of mass was about 4 cm with well-defined lesions with high signal intensity on T2WI (A) and low signal intensity on T1WI (B).

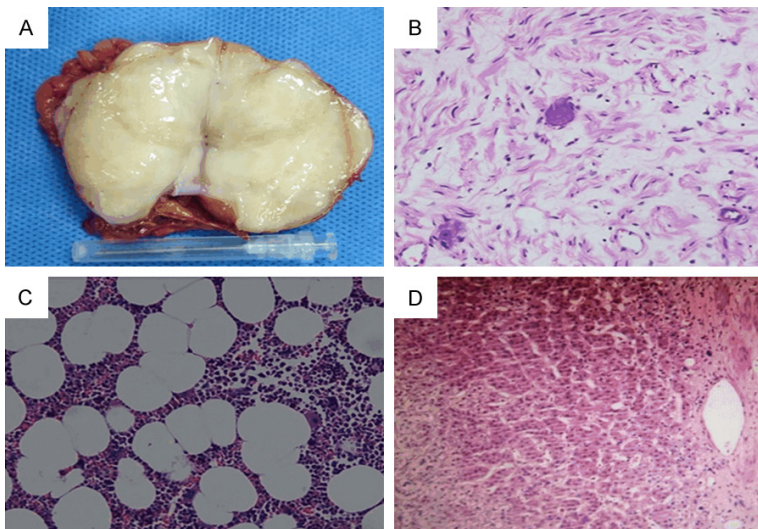


Figure 2. Histologic findings. A. Sectioned surface of adrenal gland revealing a large gray-white and firm mass with a gray-red soft area at one edge of the tumor. B. Adrenal ganglioneuroma consisted of interlacing bundles of neurofibromatous tissue with mature ganglion cells scattered throughout (200 \times). C. Mature adipose tissue mixed with hematopoietic tissue was identified (200 \times). D. This was mingled with the neurofibromatous tissue, some of which was seen in close proximity to the myelolipomatous foci (40 \times).

underwent an en-bloc resection of the mass via microlaparoscope technique after positive pre-operative preparation. The operation was successful and postoperative recovery was well.

Pathologic findings

Pathologic examination showed a 6.0*6.0*2.2 cm solid tumor with well-defined boundary enlarging the adrenal medulla. The tumor had a segmented surface, which was gray-white and firm with a gray-red soft area with a size of 2.5 cm, at one edge of the tumor (**Figure 2A**). The microscopic results showed that the tumor had an obvious boundary and had interlacing bundles of neurofibromatous tissue with scattered

mature ganglion cells (**Figure 2B**). In this tumor, there were several foci (i.e. the gray-red area through gross examination) of mature adipose tissue which were blended with hematopoietic tissue (**Figure 2C**). The majority of the foci were found in the tumor's peripheral area. Strands of adrenal cortical cells, the majority of which were reticular type, were mixed with the neurofibromatous tissue, and it was found that some of them were in close association with the myelolipomatous foci (**Figure 2D**).

Discussion

This case represents an unusual coexistence of adrenal myelolipoma related to a ganglioneuroma of adrenal medulla. The adrenal gland consists of two major components that differ in the embryologic origin. A collision tumor has been rarely seen where two neoplasms are coexisting but independent from each other and have no substantial histologic admixture at the interface. The most frequently seen collision tumor in adrenal gland is an adrenal adenoma with myelolipoma [5]. Other reported collision tumors in adrenal glands include

ganglioneuroma with adenoma, or myelolipoma and metastasis into an adenoma or myelolipoma [6, 7]. As far as we know, a myelolipoma occurring related to an adrenal ganglioneuroma was rarely reported.

Adrenal myelolipomas are rare. The benign neoplasms of the adrenal gland have different clinical presentations and the pathogenesis is still uncertain. The tumor usually presents as lipomatous to different degrees as along with myeloid tissue. Typically, a lipid tissue contains an active bone marrow component, presenting normal trilineage hematopoiesis but with a significantly increased quantity of megakaryocytes. On the other hand, the association of

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myelolipomas with hormonally active states and neoplastic conditions increases the possibility that the hormonal microenvironment may contribute to the development of myelolipomas. At the same time, the National Institute of Health Consensus Panel on adrenal incidentalomas (2002) summarized that adrenal myelolipomas can be seen as an exceptional case to the mandatory metabolic work-up of an adrenal mass that was recently discovered.

Adrenal ganglioneuroma has a prevalence ranging from 5% to 9.4% in many adrenalectomies [9]. As a result of the rarity, this type of tumor has relatively few available materials and most of them are seen in the literature. Ganglioneuromas usually appear as a homogeneous round mass with clear boundary, and often surrounding major blood vessels [10]. They recur or metastasize to the regional lymph nodes or distant organs with a low frequency [11].

Therefore, this type of adrenal tumor can be verified by pathologic examination. A careful workup combination of laboratory, endocrine, and imaging investigations is essential. If patients present with benign imaging findings and have nonspecific hormonal references, laboratory, endocrine, and imaging investigations should be repeated at 6, 12, and 24 months and they should be followed up annually for an additional 4 years [12]. Surgery should be considered when the tumor's diameter is 4 cm or more, the tumor size increases by 1 cm or more, or newly-developed hormonal symptoms or abnormal endocrine results appear [13].

In conclusion, ACT is rare. Multi-slice Spiral CT (MSCT), MRI, and other imaging techniques can help the diagnosis. MRI has a unique advantage in the diagnosis of intratumoral fat and bleeding. Through sampling, careful pathologic examination and extensive immunohistochemical investigation are often required to confirm the components of tumor. Therefore, this paper presents this distinctive case of the coexistence of myelolipoma and ganglioneuroma in the right suprarenal fossa.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Rongzhen Tao, Department of Urology, The Affiliated Jiangning

Hospital with Nanjing Medical University, Nanjing 211100, Jiangsu, China. Tel: +86-25-86863886; Fax: +86-25-83724440; E-mail: 18915973028@163.com

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