УДК 617.5-089
DOI: 10.35693/2500-1388-2020-5-4-272-277

Myxoid adrenal cortical adenoma and isolated adrenal myelolipoma

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Abstract

Aim – to reveal the clinical, morphological and hormonal characteristics of patients operated on for isolated adrenal myelolipoma and myxoid adenomas with adrenal myelolipoma.

Material and methods. The study was based on the results of examination of patients operated on for adrenal myelolipoma in the period from 2000 to 2015. During the study, the hormonal activity in patients with isolated and myxoid adrenal myelolipomas was analyzed. A pathomorphological and immunohistochemical study of the removed formations of the adrenal gland was performed. The article presents rare clinical observations of patients undergoing treatment in the clinic of E.E. Eichwald, North-Western State Medical University named after I.I. Mechnikov, with accidentally detected neoplasms, which were a combination of adenoma and myelolipoma of the adrenal gland.

Results. The occurrence of isolated myelolipomas and myxoid adenomas with myelolipoma among incidentalomas was 6.3% and 4.9%, respectively. The average age of patients with adrenal myelolipoma is 54.5 years. The ratio of women to men was 2:1. According to our data, four types of hormonal activity have been identified among myxoid formations of the adrenal gland. Type 1 is associated with Cushing's syndrome, type 2 is associated with Conn's syndrome, type 3 is associated with autonomic cortisol secretion, type 4 has no hormonal activity. Based on the results obtained by high-performance liquid chromatography (HPLC), 11β-hydroxylase deficiency was noted, and according to the study of the urine steroid profile by gas chromatography-mass spectrometry (GCMS), the signs of type 2 11β-hydroxysteroid dehydrogenase deficiency and an increase in 5α-reductase activity in patients with isolated adrenal myelolipoma were obtained.

Conclusion. The combination of adenoma with myelolipoma, as well as of adenoma, myelolipoma and cavernous hemangioma is a rare pathology, the diagnosis of which is based on the pathomorphological examination of the removed tumor. Myelolipoma as a part of adrenal adenoma is an incidentaloma, which can be of different sizes and is accompanied by hormonal dysfunction of the cells of the adrenal cortex. An uncertain intrascopic phenotype and hormonal activity of myxoid formations are the decisive factors in favor of surgical intervention.

Keywords: adrenal myelolipoma, myxoid adrenal tumor, adrenal adenoma, endovideosurgical adrenalectomy.

Conflict of interest: nothing to disclose.

Citation
Lisitsyn AA, Zemlyanoi VP, Velikanova LI, Bekhtereva IA, Shafigullina ZR. **Myxoid adrenal cortical adenoma and isolated adrenal myelolipoma.** *Science & Innovations in Medicine.* 2020;5(4):272-277.
doi: 10.35693/2500-1388-2020-5-4-272-277

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Received: 02.09.2020
Revision Received: 30.09.2020
Accepted: 01.10.2020

Миксоидная кортикальная аденома и изолированная миелолипома надпочечника

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Аннотация

Цель – провести клинико-морфологический и гормональный анализ оперированных больных с изолированными миелолипомами и миксоидными аденомами с миелолипомой надпочечника.

Материал и методы. В основу работы легли полученные результаты обследования оперированных больных с миелолипомами надпочечника за период с 2000 по 2015 г. Был проведен анализ гормональной активности изолированных и миксоидных миелолипом надпочечника. Выполнено патоморфологическое и иммуногистохимическое исследование удаленных образований надпочечника. В работе представлены редкие клинические наблюдения больных, проходивших лечение в клинике Э.Э. Эйхвальда СЗГМУ им. И.И. Мечникова со случайно выявленными образованиями, представленными комбинацией аденомы и миелолипомы надпочечника.

Результаты. Частота встречаемости изолированных миелолипом и миксоидных аденом с миелолипомой среди инциденталом составила 6,3% и 4,9% соответственно. Средний возраст больных с миелолипомой надпочечника равен 54,5 года. Соотношение между женщинами и мужчинами составило 2:1. На основании полученных данных выявлено четыре типа гормональной активности миксоидных образований надпочечника. 1 тип ассоциирован с синдромом Кушинга, 2 тип – с синдромом Конна, 3 тип – с автономной секрецией кортизола, 4 тип – без гормональной активности. На основании результатов, полученных методом высокоэффективной жидкостной хроматографии (ВЭЖХ), отмечена 11β-гидроксилазная недостаточность, а по данным исследования стероидного профиля мочи методом газовой хромато-масс-спектрометрии (ГХМС) получены признаки недостаточности 11β-гидроксистероиддегидрогеназы 2 типа и повышения активности 5α-редуктазы у больных с изолированной миелолипомой надпочечника.

Заключение. Ассоциации аденомы с миелолипомой, а также аденомы, миелолипомы и кавернозной гемангиомы являются редкой патологией, диагностика которой основана на патоморфологическом исследовании удаленной опухоли. Миелолипома в составе аденомы надпочечника является инциденталомой, которая может быть разного размера и сопровождаться гормональной дисфункцией клеток коркового слоя надпочечника. Неопределенный интраскопический фенотип и гормональная активность миксоидных образований являются решающими факторами в пользу хирургического вмешательства.

Ключевые слова: миелолипома надпочечника, миксоидная опухоль надпочечника, аденома надпочечника, эндовидеохирургическая адреналэктомия.

Конфликт интересов: не заявлен.

Для цитирования:
Лисицын А.А., Земляной В.П., Великанова Л.И., Бехтерева И.А., Шафигуллина З.Р. **Миксоидная кортикальная аденома и изолированная миелолипома надпочечника.** *Наука и инновации в медицине.* 2020;5(4):272-277.
doi: 10.35693/2500-1388-2020-5-4-272-277

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УЗИ – ультразвуковое исследование; КТ – компьютерная томография; АКТГ – адренокортикотропный гормон; ПДТ – подавляющий дексаметазоновый тест; ВЭЖХ – высокоэффективная жидкостная хроматография; СПМ – стероидный профиль мочи; ГХМС – газовая хромато-масс-спектрометрия.

Рукопись получена: 02.09.2020
Рецензия получена: 30.09.2020
Решение о публикации принято: 01.10.2020

INTRODUCTION

Adrenal myelolipoma is a benign tumor consisting of mature adipose and myeloid tissues. The incidence of myelolipomas among adrenal gland incidentalomas is 6–16% [1]. Myelolipoma is believed to be a hormonally inactive tumor and can be excluded from mandatory hormonal screening. However, approximately 18.3% of myelolipomas are associated with hormonal dysfunction, requiring extensive hormonal examinations [2, 3]. Most tumors are diagnosed accidentally during routine ultrasonographic or tomographic examinations. Based on the results of the endoscopic diagnosis, adrenal myelolipoma can be an isolated myelolipoma, or a myelolipoma with hemorrhage, or myelolipomatous foci in the adrenal tissue [4]. One of the poorly understood pathologies of the adrenal gland are myxoid tumors. A myxoid tumor is most often an accidental finding in the pathomorphological examination of the removed adrenal gland. The coexistence of a myelolipoma within an adenoma of the cortical layer of the same adrenal gland is seen rarely [5, 6]. According to the literature, this symbiosis is most often combined with the hormonal activity of the tumor, accompanied by Cushing's syndrome or Conn's syndrome [5, 7]. The presence of a myelolipoma in a non-functional adrenal adenoma is an orphan disease [8].

Analysis of patients who underwent surgery for adrenal myelolipoma should help in resolving the issues regarding the diagnostic approach and the further treatment approach for this category of patients.

AIM

The study aimed to conduct the clinical, morphological, and hormonal analysis of patients who underwent surgery for adrenal myelolipomas.

MATERIAL AND METHODS

Over a 16-year period in the E.E. Eichwald clinic of Mechnikov North Western State Medical University, 285 patients with various formations of the adrenal gland were operated. All patients underwent a lateral transperitoneal endoscopic adrenalectomy. Among 143 patients who underwent surgery for formations detected accidentally, 16 (11.2%) patients (11 women and 5 men) had myelolipoma. All patients underwent a comprehensive examination, including a contrast-enhanced computed tomography (CT) scan and an ultrasound examination of the abdominal organs. The hormonal study was conducted using the immunoassay method with the determination of the circadian rhythm of the secretion of cortisol and adrenocorticotropic hormone (ACTH), aldosterone and renin in the blood serum, free cortisol in saliva at 23 o'clock, metanephrine and normetanephrine in blood plasma, as well as the suppressive test with 1 mg dexamethasone (SDT). Additionally, using the high-performance liquid chromatography (HPLC) method, the precursors of aldosterone and cortisol in the blood serum, urinary excretion of free cortisol (UFF), free cortisone (UFE), and 18-OH-corticosterone (U18-OHB) were determined, which enabled to establish the hormonal activity of the

Hormonal indices	Mixed tumor (aden. + myelolipoma) Itsenko-Cushing syndrome n = 2 (M/F)	Mixed tumor (aden. + myelolipoma) Conn syndrome n = 1 (M)	Mixed tumor (aden. + myelolipoma). (aden. + myelolipoma + hemangioma) with autonomous production of cortisol n = 3 (F)	Mixed tumor (aden. + myelolipoma + hemangioma) without hormonal activity n = 1 (F)
Cortisol (9 o'clock). nmol/l	525/217	321	266/527/360	438
Cortisol (21 o'clock). nmol/l	527/231	182	206/284/215	201
ACTH (9 o'clock). pg/ml	<5/<5	11	<5	8
ACTH (21 o'clock). pg/ml	<5/<5	26	<5	15,1
Aldosterone. pg/ml	24,1/54,2	208	87,5/81/39,9	44,8
Aldosterone after saline sample tests. pg/ml	–	153 (в норме<100)	–	–
Renin. pg/ml	0,5/2,5	1,2	9,2/4,5/4,6	15
Aldosterone-renin ratio	48,2/21,68	173,3	9,51/18/8,7	2,98
Metanephrine.pg/ml (0–90)	9,8/60,1	18,2	32/40,6/12,8	26,7
Normetanephrine.pg/ml (0–180)	14,3/44,2	73,4	24/62,3/18,9	35,5
Cortisol after SDT with 1 mg. nmol/l (<55)	670,0/283,2	32	207/109/145	40
Cortisol after SDT with 2 mg. nmol/l (<55)	651,0/323,0 (<55)	–	77,5/–/–	–
Cortisol after SDT with 8 mg. nmol/l (< 55)	420,0/367,0	–	185/–/–	–
Free saliva cortisol at 23 o'clock. nmol/l	64,4/20,8	6,4	15/13/14	9

Таблица 1. *Содержание уровней гормонов в плазме крови у больных со смешанной опухолью надпочечника по данным иммунохемилюминесцентного анализа*
Table 1. *The hormonal levels in blood plasma in patients with myxoid adrenal tumor according to immunochemiluminescence data*

tumors. In order to search for adrenal malignancy markers and defects in adrenal steroidogenesis, the analysis of the urine steroid profile (USP) by gas chromatography-mass spectrometry (GCMS) was used. The removed adrenal gland with a tumor was subjected to post-mortem examination.

Immunohistochemical study was performed on paraffin sections. The antibodies used were CD31 and podoplanin (D-2-40). The results were evaluated using an OLYMPUS BX-46 light-optical microscope.

Statistical data processing was performed using the STATISTICA for WINDOWS software system (version 10).

RESULTS

The mean age of the patients was 54.5 ± 6.1 years. Isolated myelolipoma was diagnosed in 9 (6.3%) of the patients, and in 7 (4.9%) cases, a combination of myelolipoma with other tumors of the adrenal gland was diagnosed. In five cases, a myxoid tumor (myelolipoma in an adenoma) was diagnosed, and in two cases, a combined tumor consisting of a cortical adenoma, myelolipoma, and cavernous hemangioma was diagnosed. The tumor was located in the right adrenal gland in 9 (56.3%) cases, and in the left adrenal gland in 7 (43.7%) cases. In 12 patients, arterial hypertension was noted; 11 patients were overweight and had an impaired glucose tolerance, and 8 patients had diabetes mellitus. Four patients with isolated adrenal myelolipoma had lumbar and abdominal pains, which was the reason for the examination. Based on the results obtained, the average size of single myelolipomas according to CT data was 8.6 ± 2.5 cm. The largest tumor, consisting of myeloid cells, was 13.1 cm. The endoscopic characteristics of the isolated myelolipomas corresponded to the adipose tissue (densitometric density, 62.9 ± 17.3 HU units). In all cases, the tumor was heterogeneous. The density of myxoid formations was 21.5 ± 5.2 HU units, and the size was 4.9 ± 0.6 cm. The dimensions of the myelolipomatous formations diagnosed in the adrenal adenoma ranged from 3 to 8 mm. Hormonal dysfunction was diagnosed in 6 out of 7 patients with a mixed tumor (**Table 1**).

Autonomic cortisol secretion was the most frequent indicator of hormonal activity in the identified mixed adrenal tumor. In one case, such a combination was diagnosed in a patient with bilateral macronodular hyperplasia. In another case, a mixed tumor (cortical adenoma, myelolipoma, and cavernous hemangioma) of the right adrenal gland with autonomic cortisol secretion was diagnosed. This tumor required a differential diagnosis with an adrenocortical cancer of the adrenal gland. The size, inhomogeneity, density, heterogeneity of the lesion with areas of reduced density, and the presence of peripheral enhancement were indicative of malignancy. At the same time, according to the USP results by the GCMS method, no data suggestive of the malignant potential were revealed. During hormonal screening, an autonomous secretion was diagnosed. Due to the indeterminate endoscopic phenotype and hormonal dysfunction, the patient was operated on a scheduled basis.

The combination of focal changes, represented by adipose tissue fragments with erythroid and granulocytic islets in solitary aldosterone-secreting adenoma and corticosteroma, was diagnosed in only three cases.

The hormone levels obtained by the immunoassay method in patients with isolated myelolipomas did not differ from those of healthy patients ($p > 0.05$) (**Table 2**). The cortisol level after SDT with 1 mg was 34.8 ± 6.1 nmol/L.

Concurrently, according to HPLC data, the level of 11-deoxycorticosterone in the blood serum increased in comparison with the indicators of hormone-inactive adenoma patients, which may be associated

Indicators	Me (LQ-UQ)	
	Patients with isolated myelolipomas (n = 9)	Healthy patients (n = 15)
ACTH at 9 o'clock, pg/ml	14,6 (13,2–18,6)	28,1 (21,2–39,3)
Cortisol at 9 o'clock, nmol/l	304 (284–375)	357 (321–426)
Cortisol at 21 o'clock, nmol/l	109 (86–172)	129 (101–203)
Free cortisol in saliva at 23 o'clock, nmol/l	8,1 (6,4–12,4)	6,5 (4,2–9,3)
Aldosterone, pg/ml	45 (35–60)	71 (45–95)
Renin, pg/ml	5,3 (2,7–41,7)	10,2 (7,1–16,2)
Aldosterone-renin ratio	5,7 (2,0–23,1)	4,4 (3,3–12,6)
Metanephrine, pg/ml	13,8 (10,5–25,2)	21,5 (15,2–48,3)
Normetanephrine, pg/ml	28,3 (15,5–45,4)	47,3 (26,2–65,2)

Таблица 2. *Показатели гормонального фона у пациентов с одиночными миелолипомами, полученные методом иммуноанализа*
Table 2. *The hormone levels in patients with single myelolipomas obtained by immunoassay*

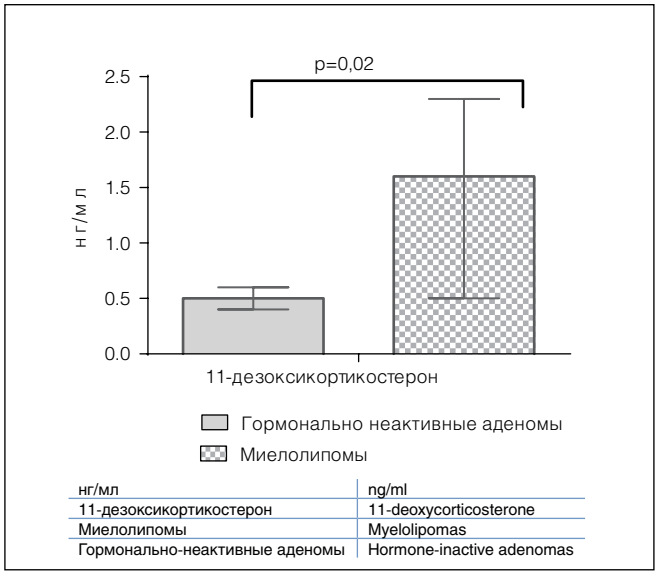

Рисунок 1. *Уровень 11-дезоксикортикостерона в сыворотке крови у больных с миелолипомой по данным ВЭЖХ.*
Figure 1. *The level of 11-deoxycorticosterone in blood serum of patients with myelolipoma according to HPLC data.*

with a deficiency of 11-β-hydroxylase in patients with solitary myelolipomas (**Figure 1**). According to GCMS data, an increase in the (THF + allo-THF)/THE and THB/THA ratios in comparison with the control group may indicate a type 2 11-β-hydroxysteroid dehydrogenase deficiency, and an increase in the urinary excretion of 11-hydroxyandrosterone and the ratio of this androgen to 11-hydroxyethiocholanolone may be associated with an increase in 5α-reductase activity (**Figure 2**).

In all cases, transperitoneal endovideosurgical adrenalectomy was performed. The duration of the surgery was 100 ± 24.1 min for the removal of solitary myelolipomas, and 90.5 ± 16.1 min for mixed tumors. Intraoperative blood loss was 22.1 ± 4.9 ml and 19.2 ± 8.6 ml, respectively. No Intra- and postoperative complications were recorded. During the follow-up period (1–12 years), there were no long-term complications or local recurrence.

DISCUSSION

Adrenal gland myelolipoma is a benign and hormonally inactive tumor consisting of mature adipose and hematopoietic tissues.

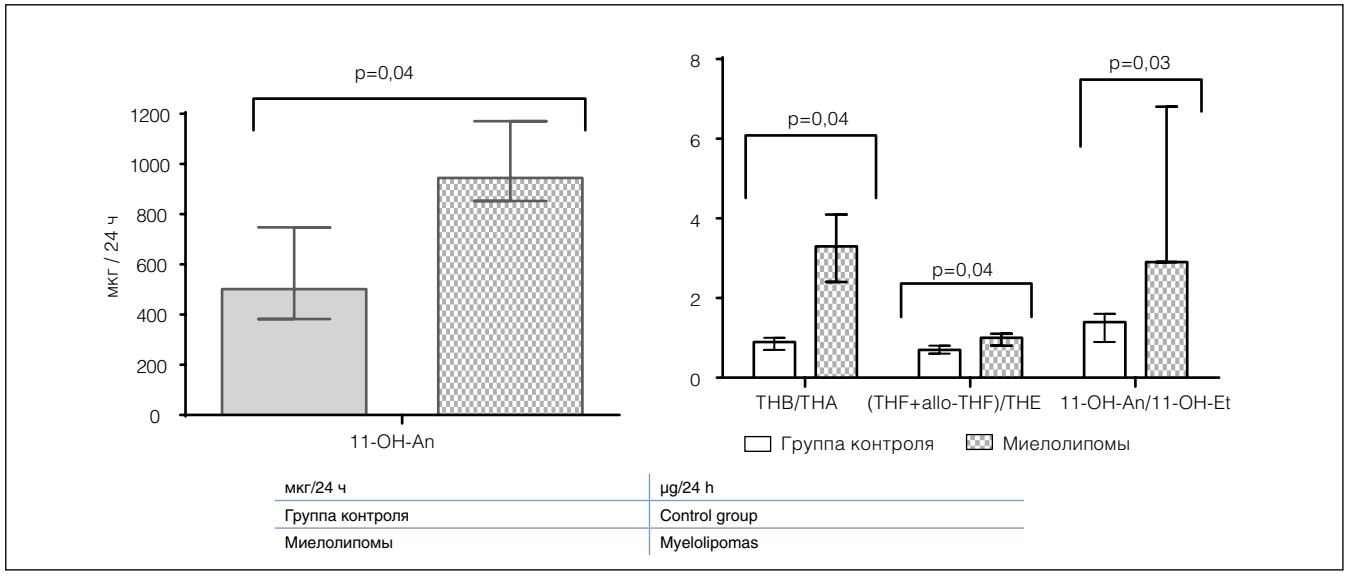

Рисунок 2. *Экскреция с мочой 11-гидроксиандростерона (11-OH-An – 5α-метаболита андростендиона) и признаки снижения активности 11β-НСД 2 типа (соотношение THB (тетрагидрокортикостерона) / THA (тетрагидро-11-дегидрокортикостерона; THF (тетрагидрокортизол)+alloTHF/THE (тетрагидрокортизон) и увеличения активности 5α-редуктазы (11-OH-An (11-гидроксиандростерон) / 11-OH-Et (11-гидроксиэтиохоланолон) у больных с миелолипомой по данным газовой хромато-масс-спектрометрии.*
Figure 2. *Urinary excretion of 11-hydroxyandrosterone (11-OH – An-5α-Androstenedione metabolite) and signs of decreased activity of 11β-HSD type 2 (ratio THB (tetrahydrocorticosterone) / TNA (tetrahydro-11-dehydrocorticosterone; THF (tetrahydrocortisol)+alloTHF/THE (tetrahydrocortisone) and increase in 5α-reductase activity (11-OH-An (11-hydroxyandrosterone) / 11-OH-Et (11-hydroxyethiocholanolone) in patients with myelolipoma according to gas chromatography-mass spectrometry.*

Currently, there are a number of theories that seek to explain the cause of this tumor. The theories include the cellular activity of hematopoietic cells remaining in the adrenal gland from the embryonic period, the transfer of emboli from the bone marrow to the adrenal tissue, and metaplasia of the adrenal reticuloendothelial cells into adipose and hematopoietic cells in response to stimuli such as stress, infection, or tissue necrosis [5]. In our cases, eight patients had anamnestic aspects (stroke, closed craniocerebral trauma, chest injury, comminuted fracture of the right humerus, detachment of the meniscus of the left lower extremity, and history of multiple surgeries), which, according to one of the theories, could be a trigger of this disease [9]. In two cases, a combination with kidney angiomyelolipoma was noted. Concurrently, three patients did not have a single disease that could serve as a trigger for this process.

There is another opinion concerning the nature of myelolipoma. This is a theory of hormonal imbalance based on experimental studies by the combined administration of an extract of the anterior pituitary gland into the adrenal tissue of gray mice, which subsequently triggered the myelolipomatous transformation of adrenal cortex cells [2, 10]. As a rule, such asymptomatic tumors are diagnosed accidentally. However, in 10% of cases, they are associated with hormonal dysfunction and are combined with Cushing's syndrome, Conn's syndrome, and congenital adrenal hypoplasia with 21-hydroxylase deficiency [11]. In our cases, hormonal activity was diagnosed in 6 out of 7 patients with myxoid tumor (autonomous production of cortisol in three patients, corticosteroma in two patients, and aldosterone-secreting adenoma in one patient). In patients with isolated myelolipoma, according to the HPLC results, hormonal dysfunction associated with an increase in the level of 11-deoxycorticosterone was noted, and according to GCMS, signs of decreased activity of type 2 11-β-hydroxysteroid dehydrogenase (11β-HSD) and an increase in the activity of 5α-reductase were revealed. In the material presented, the incidence of myxoid formations by gender was (F/M = 4/3), and that of isolated myelolipoma tumor was (F/M = 6/3). The ratio of the lesion side of the mixed formations was 3 on the right and 4 on the left, and that of solitary myelolipomas was 6 on the right and 3 on the left.

Due to the asymptomatic course, the diagnosis and indications for surgery are based on the CT data obtained. Myelolipoma is a hypodense (from –30 to –90 HU), heterogeneous tumor with clearly distinct contours.

However, in some cases, the diagnosis of isolated myelolipoma requires special attention. It should be differentiated from liposarcoma, extrarenal angiomyelolipoma, leiomyosarcoma, myxofibrosarcoma, teratoma, and adrenocortical carcinoma [11]. X-ray diagnosis of myxoid tumor (myelolipoma in adenoma) is not always possible. In our study, a combination of the tumor with myelolipoma was suspected in only two cases during endoscopic screening. In all other cases, this was an incidental finding. Based on the CT data obtained, adrenocortical cancer had to be ruled out in two patients with composite tumor. In our study, all patients underwent surgery because of the uncertainty of the endoscopic phenotype, size, and heterogeneity of the tumor diagnosed. Steroid urine profile by GCMS did not reveal any markers of adrenocortical cancer. However, the final diagnosis in all cases was established on the basis of histological and immunohistochemical studies. None of the pathological conclusions provided evidence for malignancy (vascular invasion, capsular invasion, mitotic activity, and necrosis). According to the L.M. Weiss scale, only in three cases of myxoid tumor was one of the criteria noted, namely the content of light cells was more than 25%.

Questions regarding the indications for surgery in patients with adrenal myelolipoma remain the subject of discussion. Some experts recommend adrenalectomy for tumors >7 cm [12]. Other authors, given the high risk of the spontaneous rupture of the myelolipoma, suggest removing the tumor ≥ 6 cm [13]. It is believed that the indication for the removal of the gland is the presence of a large tumor, the uncertainty of the endoscopic diagnosis, and the presence of clinical symptoms [4]. At the same time, the American Association of Clinical Endocrinology and the American Association of Endocrine Surgeons in 2009 recommend performing adrenalectomy in patients with myelolipomas >4 cm [14]. Among the operated patients in our clinic, the removed isolated myelolipomas were 5–13 cm in size.

Endovideosurgical adrenalectomy is a safe and effective method. Currently, two approaches are used to remove adrenal tumors, namely transperitoneal and retroperitoneal. Although the transperitoneal access is most convenient for large lesions, none of the methods have significant advantages. Possible organ-sparing surgeries in adrenal myelolipomas have been suggested. Thus, a group of specialists proposes to remove myelolipoma by retroperitoneal liposuction. This approach allows not only to minimize surgical trauma, but it also ensures a relapse-free surgical intervention [15]. Others suggest removing the tumor while sparing the adrenal cortex [16]. However, despite the low risk of the myelolipoma-carcinoma association, uncertain CT characteristics, and large tumor sizes, we performed the complete removal of the adrenal gland with the lesion. Also, all tumors in which the combination of adenoma with myelolipoma was subsequently diagnosed were removed together with the adrenal gland. Despite the large size of myelolipomas, endovideosurgical adrenalectomy was performed. Criteria such as benign nature, mobility of the tumor during its isolation, and a low percentage of local recurrence offer preferable conditions for the endovideosurgical removal of the adrenal gland with the tumor. This approach is technically feasible and safe in the treatment of large myelolipomatous formations of the adrenal gland.

CONCLUSIONS

1. The association of adenoma with myelolipoma is a rare pathology, whose diagnosis is based on the pathomorphological examination of the tumor removed.

2. Myelolipoma as part of an adrenal adenoma is an incidentaloma, which can be of different sizes and can be accompanied by hormonal dysfunction of the adrenal cortex cells. Uncertain endoscopic phenotype and hormonal activity of myxoid formations are decisive factors in favor of a surgical intervention.

3. Patients diagnosed with isolated adrenal myelolipomas require mandatory hormonal screening.

Conflict of interest. *The authors declare no conflict of interest.*

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