Case Report

Giant symptomatic adrenal myelolipoma: A case report

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A R T I C L E   I N F O

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A B S T R A C T

Introduction: Adrenal myelolipomas are rare non-functioning benign tumors composed of adipose and hematopoietic tissues. Most AMLs are discovered incidentally and represent the second most common adrenal incidentaloma.

Case presentation: A 58-years-old female patient, obese with a history of diabetes and blood hypertension presented with complaints of pain in the left flank. Abdominopelvic computed tomography showed a giant well-defined mass of the left adrenal gland with fat density suggesting adrenal myelolipoma. The patient underwent open left adrenalectomy. The pathological study confirmed the diagnosis of adrenal myelolipoma.

Discussion: Most AMLs are asymptomatic, remain stable in size, or grow slowly. Mass effect symptoms and spontaneous rupture are observed more in larger AMLs. The most common symptoms observed are abdominal discomfort/pain, hypochondrial pain, and flank pain. Most of the AMLs are discovered incidentally and the radiological features are accurate in diagnosing AML in up to 90% of the cases, CT is more sensitive for detection than other imaging modalities. The open surgery approach is the standard treatment of choice for giant AML (>10cm) while the minimally invasive approach has been used in only a few cases.

Conclusion: The therapeutic management is discussed on a case-by-case basis. Surgical treatment is indicated for larger, symptomatic, or rapidly growing AMLs. Meanwhile smaller and asymptomatic AMLs are managed conservatively.

1. Introduction

Adrenal myelolipomas (AMLs) are rare non-functioning benign tumors composed of adipose and hematopoietic tissues [1]. AMLs are found in one out of 500–1250 autopsy cases [2]. However, the exact clinical prevalence of the tumor is impossible to assess because of the high percentage of asymptomatic cases and its benign nature [2]. Most AMLs are asymptomatic and discovered incidentally; they represent the second most common adrenal incidentaloma with 6–16% of all incidental adrenal masses [1, 2]. The Radiological features are specific and suggest the diagnosis in more than 90% of the cases [3]. Myelolipoma is defined as ‘giant’ when its greatest diameter is >10 cm [4]. Giant AMLs are exceptional and reported only in a few studies Herein, we report a rare case of a giant symptomatic left AML in a female patient. This case has been reported following the SCARE criteria [5].

2. Case report

A 58-years-old female patient, obese (BMI: 35Kg/m²) with a history of diabetes and blood hypertension under treatment with no endocrine disorders or associated comorbidities in the patient and relatives, presented with a three months history of vague, dull pain in the left flank without fever or urinary signs. Physical examination revealed a slight tenderness in the left lumbar region without any palpable mass. Abdominal ultrasound showed the presence of a hyperechoic mass in the left suprarenal region with undefined margins. An abdominopelvic computed tomography (CT) showed a large well-defined mass of the left adrenal gland measuring 111 × 82 × 82 mm with fat density (~80 UH) suggesting an adrenal myelolipoma (Fig. 1). Laboratory tests, including routine blood examination, serum cortisol 8 a.m., 24-h urine cortisol excretion, Urinary normetanephrine, and metanephrine, were all within the normal range.

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The patient underwent open left adrenalectomy. The specimen size was $11 \times 13 \times 6$ cm and weighed 400 g (Fig. 2); Pathology examination showed the presence of fat and hematopoietic tissues confirming the diagnosis of adrenal myelolipoma (Fig. 3). The patient made an uneventful recovery and was discharged from the hospital on postoperative day five. She remained asymptomatic at a one-year follow-up.

3. Discussion

Described for the first time by Gierke in 1905 [6]. AML tends to occur more frequently in the fifth and sixth decades with a mean age of 51 years without any gender predominance [2]. According to a large study, 73% of the patient suffered from hypertension and more than half of the patients were obese with BMI $>30$ kg/m$^2$ [7]. According to Decmann et al., 59.2% of the AML reported in their review were located in the right adrenal gland and only 12.3% were bilateral [2]. In a large study conducted by Hamidi et al., only 11 patients among 305 had AML $\geq 10$ cm [1]; the largest reported AML in the literature was measuring $31 \times 24.5 \times 11.5$ cm and weighing 6 kg [8]. Myelolipomas remain stable in size or grow slowly, in a large longitudinal follow-up study, overall tumor change ranged from $-10$ mm to 115 mm with growth rate ranging from $-6$ mm/year to 14 mm/year [1].

Most AMLs are asymptomatic, mass effect symptoms and spontaneous rupture are observed more in AML $>6$ cm [1,9]. The most common symptoms observed are abdominal discomfort/pain, hypochondrial pain, and flank pain [2]. Most of the AMLs are discovered incidentally and the radiological features are accurate in diagnosing AML in up to 90% of the cases [9], ultrasound is not accurate and may show hypoechoic or hyperechoic mass depending on the predominance of fat or myeloid tissue [10]. CT is more sensitive for detection than other imaging modalities [11], AMLs appear well-defined, hypodense, and heterogeneous masses; the presence of fat density is essential for the radiological diagnosis of AML [2]. On magnetic resonance imaging (MRI), fat tissue demonstrates high-intensity signal on T1-weighted.
several cases [17, 20]. In our case, open left adrenalectomy was performed as the tumor was giant and symptomatic. Table 2 illustrates the management of giant AMLs with the outcome and follow-up.

4. Conclusion

Most AMLs are asymptomatic and discovered incidentally. The radiological findings are accurate and make the diagnosis in more than 90% of cases. The therapeutic management is discussed on a case-by-case basis. Surgical treatment is indicated for larger, symptomatic, or rapidly growing AMLs meanwhile smaller and asymptomatic AMLs are managed conservatively.

Ethics approval

No ethical approval necessary.

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Author contributions

Ramdani Abdelbassir: Writing, review and editing of the manuscript.

Asmae Aissaoui, Amal Bennani: Provided the pathological analysis.

Bouhout Tariq, Latrach Hanane, Serji Badr: Contributed for diagnosis and treatment of the patient.

El Harroudi Tijani: Supervised the writing of manuscript.
Registration of research studies

Our paper is a case report; no registration was done for it.

Guarantor

Ramdani Abdelbassir.

Consent of the patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

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Declaration of competing interest

The authors declared no potential conflicts of interests with respect to research, authorship and/or publication of the article.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103333.

References

[1] O. Hamidi, R. Raman, N. Lazik, et al., Clinical course of adrenal myelolipoma: a long-term longitudinal follow-up study, Clin. Endocrinol. 93 (1) (2020) 11–18, https://doi.org/10.1111/cen.14188.
[2] Á. Decmann, P. Perge, M. Tóth, P. Igaz, Adrenal myelolipoma: a comprehensive review, Endocrine 59 (1) (2018) 7–15, https://doi.org/10.1007/s12020-017-1473-4.
[3] Y. Nakayama, N. Matayoshi, M. Akiyama, et al., Giant adrenal myelolipoma in a patient without endocrine disorder: a case report and a review of the literature, Case Rep Surg 2018 (2018) 4854368, https://doi.org/10.1155/2018/4854368. Published 2018 Jun 11.
[4] M.R. Bokhari, S. Bhimji, Adrenal myelolipoma. StatPearls NCBI Bookshelf, 2017.
[5] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical Case Report (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[6] E. Gierke, Uber knochenmarksgewebe in den nebenniere, Betin. Path. Anat. 7 (1005) 311–325.
[7] M.J. Campbell, M. Obasi, B. Wu, M.T. Corwin, G. Fananapazir, The radiographically diagnosed adrenal myelolipoma: what do we really know? Endocrine 58 (2) (2017) 289–294, https://doi.org/10.1007/s12020-017-1410-6.
[8] H. Akamatsu, M. Koseki, H. Nakaba, et al., Giant adrenal myelolipoma: report of a case, Surg. Today 34 (2004) 283–285, https://doi.org/10.1016/S0969-5004(03)00224-4.
[9] V.G. Shenoy, A. Thota, R. Shankar, M.G. Desai, Adrenal myelolipoma: controversies in its management, Indian J. Urol 31 (2) (2015) 94–101, https://doi.org/10.4103/0970-1591.152807.
[10] S. Adapa, S. Naramala, V. Gayam, et al., Adrenal incidentaloma: challenges in diagnosing adrenal myelolipoma, J Investig Med High Impact Case Rep. 7 (2019), https://doi.org/10.1177/232470961870311, 232470961870311.
[11] S. Kumar, K. Jayant, S. Prasad, et al., Rare adrenal gland emergencies: a case series of giant myelolipoma presenting with massive hemorrhage and abscess, Nephro-Urol. Mon. 7 (1) (2015), e22671, https://doi.org/10.5812/numonthly.22671. Published 2015 Jan 19.
[12] K.M. Elsayes, G. Mukundan, V.R. Narra, et al., Adrenal masses: mr imaging features with histopathologic correlation, Radiographics 24 (Suppl 1) (2004) S73–S86, https://doi.org/10.1148/rg.24si045514.
[13] Y.K. Gao, Z.G. Yang, Y. Li, et al., Uncommon adrenal masses: CT and MRI features with histopathologic correlation, Eur. J. Radiol. 62 (3) (2007) 359–370, https://doi.org/10.1016/j.ejrad.2006.12.011.
[14] M.A. Zeiger, G.B. Thompson, Q.Y. Duh, et al., American association of clinical endocrinologists and American association of endocrine surgeons medical guidelines for the management of adrenal incidentalomas: executive summary of recommendations, Endocr. Pract. 15 (5) (2009) 450–453, https://doi.org/10.4158/EP.15.5.450.
[15] S.W. Hsu, K. Shu, W.C. Lee, Y.T. Cheng, P.H. Chiang, Adrenal myelolipoma: a 10-year single-center experience and literature review, Kaohsiung J. Med. Sci. 28 (7) (2012) 377–382, https://doi.org/10.1016/j.kjms.2012.02.005.
[16] R.A. Gadellareem, A.M. Moen, M. Khalil, et al., Experience of a tertiary-level urology center in clinical urological events of rare and very rare incidence. V. Urological tumors: 1. Adrenal myelolipoma, Curr Urol 14 (2) (2020) 80–91, https://doi.org/10.1159/000499254.
[17] G. Cochetti, A. Paladini, A. Boni, et al., Robotic treatment of giant adrenal myelolipoma: a case report and review of the literature, Mol Clin Oncol 10 (5) (2019) 492–496, https://doi.org/10.3892/mco.2019.1823.
[18] M. Ramirez, S. Miura, Adrenal myelolipoma: to operate or not? A case report and review of the literature, Int J. Surg. Case. Rep. 5 (8) (2014) 494–496, https://doi.org/10.1016/j.ijscr.2014.04.001.
[19] A.K. Lam, Update on adrenal tumours in 2017 World Health Organization (WHO) of endocrine tumours, Endocr. Pathol. 28 (2017) 215–227.
[20] X.L. Tso, S.K. Lim, Robotic assisted adrenalectomy: is it ready for prime time? Invest. Clin. Urol. 57 (Suppl 2) (2016) S130–S146.