Laparoscopic adrenalectomy via transperitoneal and retroperitoneal approaches for bilateral giant adrenal myelolipoma: A case report and literature review

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Abstract. Adrenal myelolipoma (AML) is a rare, benign neoplasm of the adrenal gland, typically asymptomatic and non-functioning. It is often detected incidentally during health checkups using B-ultrasound or computed tomography scans. Bilateral giant AMLs treated with combined abdominal and retroperitoneal laparoscopic adrenalectomy are rare. The present study reports on a 60-year-old male incidentally diagnosed with bilateral giant adrenal masses measuring 100x87x63 mm on the right and 75x52x33 mm on the left. The tumors were successfully excised using transperitoneal and retroperitoneal laparoscopic approaches, respectively. Histopathological examination confirmed the diagnosis of AML. The patient had an uneventful postoperative recovery and no signs of recurrence were observed during a 7-month outpatient follow-up. Although open radical adrenalectomy remains the standard treatment for giant AMLs (>6 cm), laparoscopic surgery offers distinct advantages, including reduced intraoperative bleeding, fewer postoperative complications and faster recovery. It may be suggested that both transabdominal and retroperitoneal laparoscopic approaches are safe and effective for giant AML resection. The choice of surgical method should be individualized based on tumor size, anatomical location and the surgeon's experience.

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Introduction

Adrenal myelolipoma (AML) is a rare, non-functioning, benign tumor originating from adrenal tissue. Composed primarily of mature adipose and hematopoietic tissue, it was first described by Gierke in 1905 (1,2). AMLs are usually asymptomatic due to their benign nature and lack of hormonal activity, unless they grow large enough to compress surrounding structures or cause spontaneous hemorrhage. Symptoms, when present, may include nonspecific abdominal or flank pain, hypertension, nausea, vomiting or hematuria (3). AMLs are most commonly detected incidentally during imaging for unrelated conditions or routine health checkups. Giant AMLs are uncommon in clinical practice and bilateral cases are exceptionally rare, often presenting significant therapeutic challenges because of the large tumor size, increased risk of hemorrhage and complex anatomical relationships with adjacent organs (4). At present, treatment options for AMLs primarily include observation or surgical intervention. While open adrenalectomy remains the gold standard for giant AMLs, laparoscopic techniques have been increasingly adopted for unilateral cases. However, there is a paucity of literature on the laparoscopic management of bilateral giant AMLs. The present study reported on a rare case of bilateral giant AML resected via transabdominal and retroperitoneal laparoscopic adrenalectomy, offering clinical insights into surgical strategy and approach selection.

Case report

A 60-year-old man with no clinical symptoms was referred to Zhejiang Provincial Hospital of Chinese Medicine (Hangzhou, China) in July 2022, after bilateral adrenal masses were detected incidentally on B-ultrasound examination. The patient had a 2-year history of hypertension (up to 170/110 mmHg) but was not on any regular antihypertensive therapy. Physical examination on first admission revealed normal vital signs, with no palpable abdominal wall mass or tenderness, and no relevant family history of genetic diseases. Laboratory tests, including adrenal hormones (catecholamines, aldosterone, adrenocorticotropic hormone, etc.), were within normal limits.

Adrenal contrast-enhanced computed tomography (CT) revealed bilateral giant adrenal masses-100x87x63 mm on the right and 75x52x33 mm on the left-with smooth margins and heterogeneous density (-35 to 44 HU) (Fig. 1). Adrenal contrast-enhanced magnetic resonance imaging (MRI) further confirmed the CT findings (Fig. 2). The patient first underwent retroperitoneal laparoscopic left adrenalectomy (Fig. 3A), followed by transperitoneal laparoscopic right adrenalectomy 1 month later (Fig. 3B). Due to liver and intestinal interference, two additional optical trocars were inserted during the right-side transperitoneal approach. The operation time for the two operations was 110 min (left) and 85 min (right), and estimated blood loss was 170 ml (left) and 150 ml (right). The two operations in succession were completed with no perioperative complications. Prophylactic steroid replacement therapy was administered after the second surgery, consisting of intravenous hydrocortisone 200 mg/day for 48 h, followed by tapering and transition to long-term oral maintenance therapy with hydrocortisone 20 mg/day and fludrocortisone 0.1 mg/day. Gross examination revealed well-encapsulated masses composed of yellow adipose and brown hematopoietic tissue (Figs. 4A and B and 5). Postoperative histopathology (H&E staining) performed according to standard procedures confirmed AML in both adrenal glands (Fig. 6). At the 3- and 6-month follow-ups, no signs of recurrence were observed via ultrasound and the patient recovered well and remained symptom-free over a 3-year postoperative period.

Discussion

AML is a rare, benign, hormonally inactive adrenal tumor, accounting for 3.3-6.5% of all adrenal masses (1). Approximately 85-90% of AMLs are detected incidentally (5,6). While AMLs are typically unilateral, ~12% are bilateral (7). According to statistics, AMLs are three times more common on the right adrenal gland than on the left (3). A long-term longitudinal follow-up study suggested that larger AMLs (>6 cm) are more often associated with bilateral involvement (6). The prevalence of AML has been reported to be 0.24% in patients undergoing CT (8), and ~0.32% in the general population at around 40 years of age (9), whereas patients with congenital adrenal hyperplasia (CAH) demonstrate a much higher rate (10.1%) (9). Additionally, bilateral or giant AMLs are more frequently observed in patients with CAH (10) and may even serve as an early sign of CAH (7). In a recent epidemiological survey, AML was detected in 4-10% of patients with adrenalectomy (9). Diagnosis of benign adrenal tumors, such as AML, is gradually becoming more accurate. Certain scholars have reported cases of adrenalectomy due to misdiagnosis (11). In most histopathological reports, the pathological features of bilateral AMLs are the same (7,12,13). Yet, the possibility of varying histopathological types of AML in the left and right adrenal glands cannot be clinically excluded to reduce the probability of misdiagnosis. The present case is a bilateral AML with the same pathological type without abnormal and correlated laboratory indicators and a disease history of CAH.

AML usually results from the differentiation of adrenal medulla or cortical cells into adipose tissue and extramedullary hematopoietic tissue. Chronic stress, infection, tissue ischemia, necrosis, trauma and fat metabolism disorders are known to cause this disease (14,15). Studies have suggested links between the adrenocorticotropic hormone, erythropoietin and AML development (6,9). Scholars have discovered the first AML-associated somatic genetic aberration in the early stage, balanced translocation between chromosomes 3q25 and 21p11, thus offering the earliest genetic evidence for tumor origin (16). Later, cytogenetic studies found that AML is a clonal tumor based on non-random X chromosome inactivation; such inactivation in hematopoietic cells and adipocytes may be of clonal origin (17). Hypotheses regarding AML pathogenesis are as follows: i) AML may be caused by an ectopic proliferation of bone marrow cells in the embryonic adrenal gland. It may originate from fetal bone marrow residues, bone marrow cell embolism and ectopic reticulocyte proliferation (11); ii) adipocytes develop from mesenchymal stem cells in the endothelium (18); iii) microRNA profiles unique to AML have been determined, distinguishing it from other adrenocortical tumors (19); and iv) scholars have found a non-pathogenic ARMC5 allelic variation in a patient with AML with a co-existing adrenal cortical adenoma. However, ARMC5 mutation is typically associated with the pathogenesis of primary bilateral macronodular hyperplasia and its correlation with myelolipoma or mixed tumors remains unelucidated (11). Bilateral AMLs may involve more complex pathogenic mechanisms than unilateral ones.

In recent years, the AML detection rate has significantly increased, largely due to the widespread use and advancement of imaging modalities such as B-ultrasound and CT. B-ultrasound, being non-invasive and radiation-free, is ideal for initial screening and follow-up monitoring of AML. However, CT remains the most accurate tool for identifying and characterizing AML, with its hallmark features being low-density fat and iso- to hyperdense marrow-like components (20). On CT, AML typically appears as a round or ovoid mass with a well-defined margin in the adrenal region. The tumor was demonstrated to have heterogeneous density, containing areas of mature fat (CT value <-30 HU) interspersed with fine reticular, cord-like or lamellar hematopoietic tissue of slightly higher density (21). Multiplanar CT reconstruction is particularly valuable in localizing lesions and evaluating their relationship with adjacent structures such as the liver and kidneys. On unenhanced MRI, AMLs with a high fat content show high signal intensity on both T1- and T2-weighted images (21). Post-contrast MRI typically shows no significant signal change due to the fat-rich nature of the tumor. The drop in the fat suppression sequence signal is characteristic of this disease and aids in diagnosis. MRI is able to more accurately display cystic changes in the tumor. Due to its multiplanar capabilities, MRI serves as a useful complement to CT in assessing tumor location and its spatial relationships with nearby organs, particularly in retroperitoneal lesions (22). When AMLs are large, intratumoral hemorrhage may occur and CT is particularly sensitive for detecting such bleeding events (23). CT is also more effective than other modalities in revealing the presence of bone marrow tissue and calcifications within the mass (24). Despite the utility of imaging, preoperative fine-needle aspiration or biopsy guided by ultrasound or CT is not generally recommended due to its invasiveness and associated risks, including

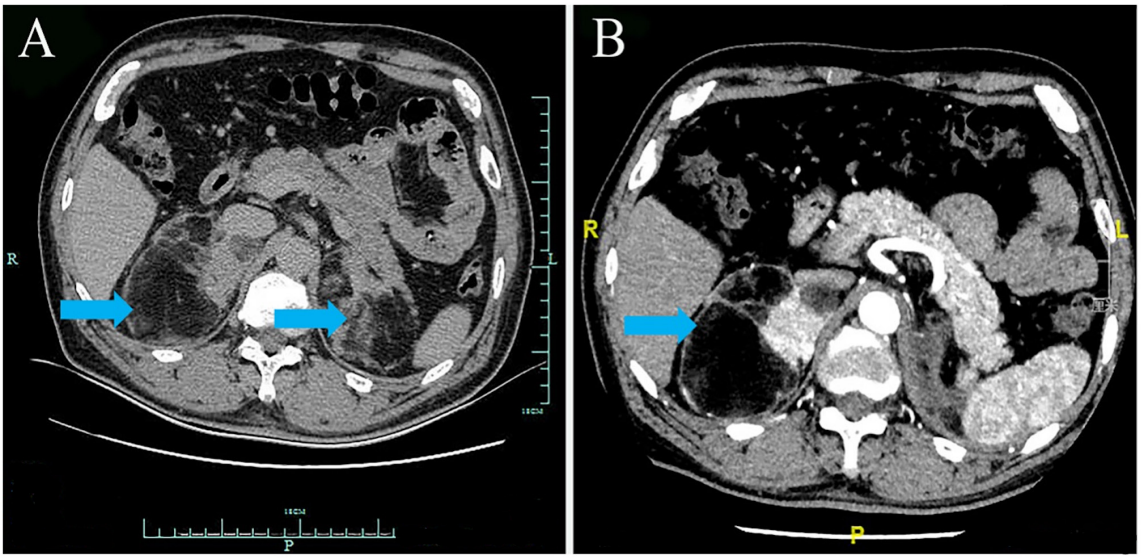

Figure 1. (A) Preoperative computed tomography displaying bilateral giant adrenal myelolipomas. (B) Contrast-enhanced CT displaying giant right adrenal myelolipoma and adrenal glands after the left-side operation (myelolipomas indicated by arrows).

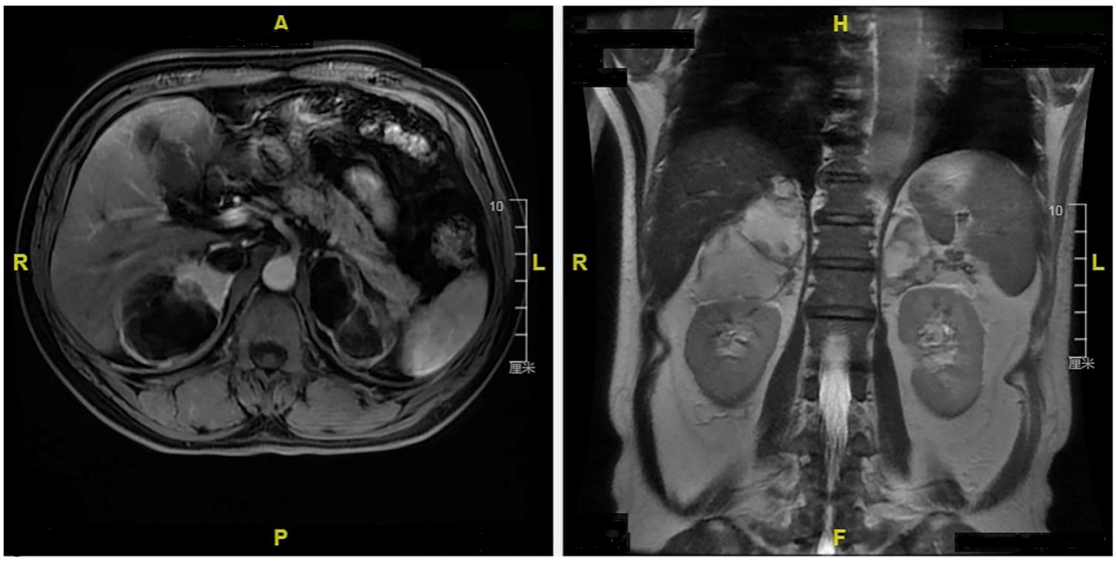

Figure 2. Horizontal and coronal abdominal magnetic resonance imaging scan showing bilateral giant adrenal myelolipoma.

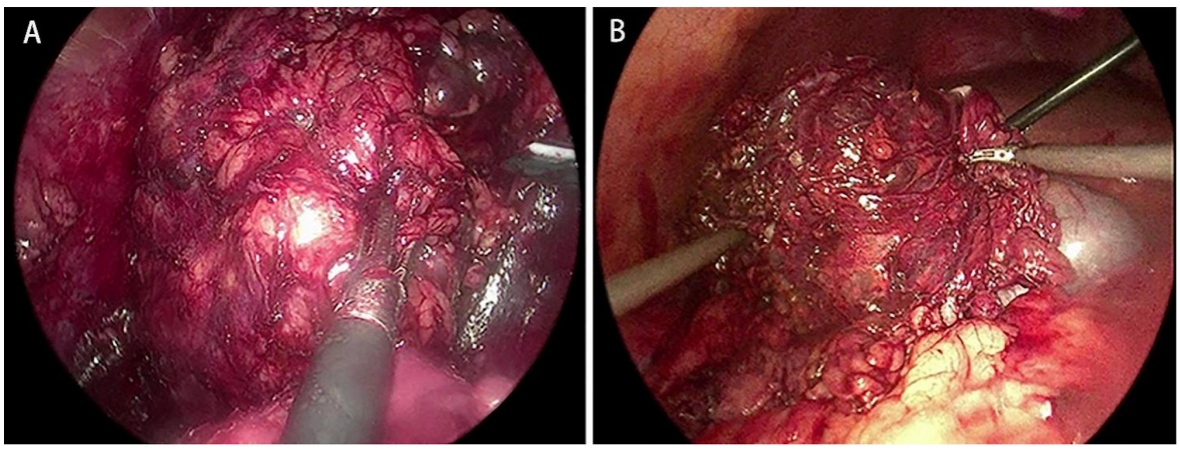

Figure 3. (A) Intraoperative image exhibiting the resected giant myelolipoma during retroperitoneal laparoscopic left adrenalectomy. (B) The resected giant myelolipoma during transperitoneal laparoscopic right adrenalectomy.

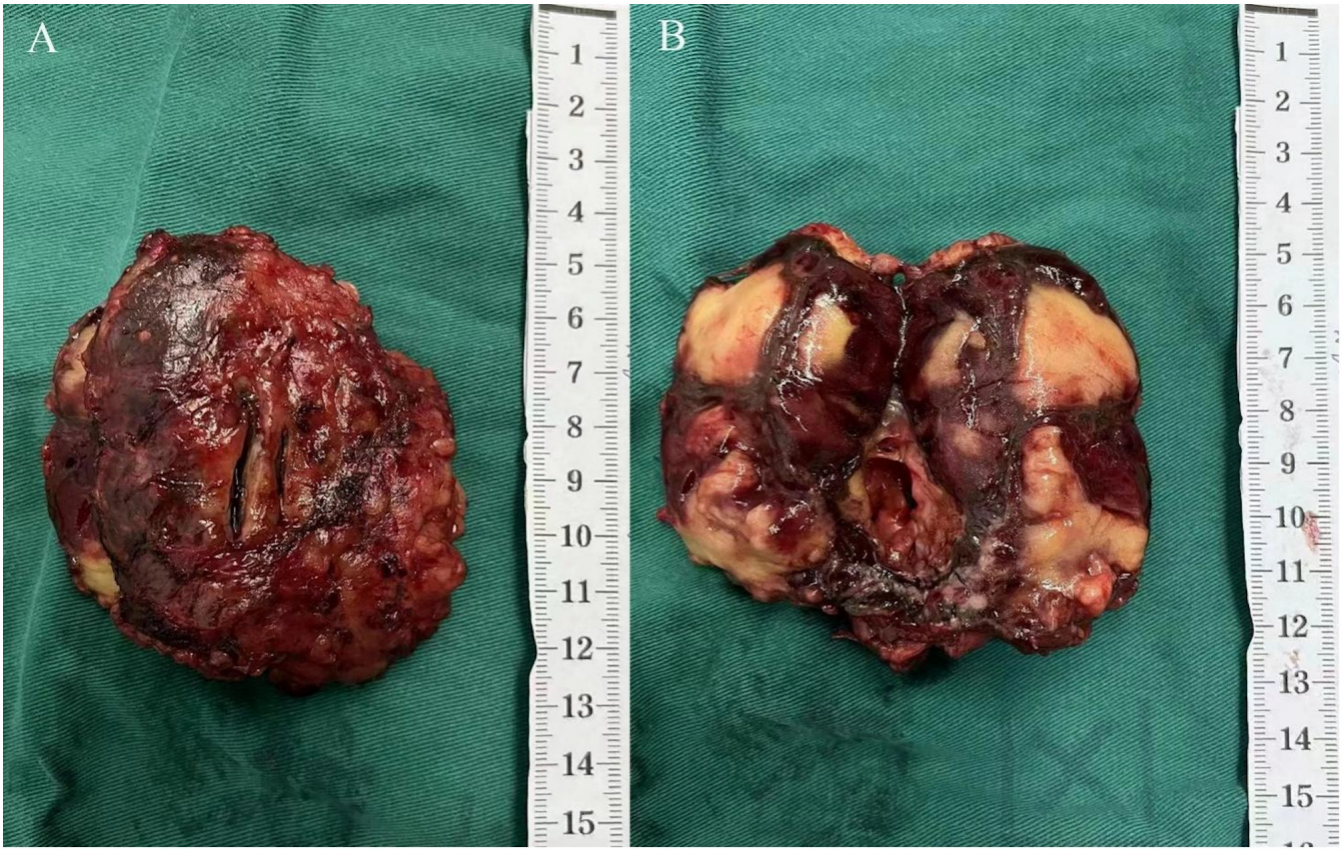

Figure 4. (A) Macroscopic view and (B) cross-sectional view of the resected giant left myelolipoma.

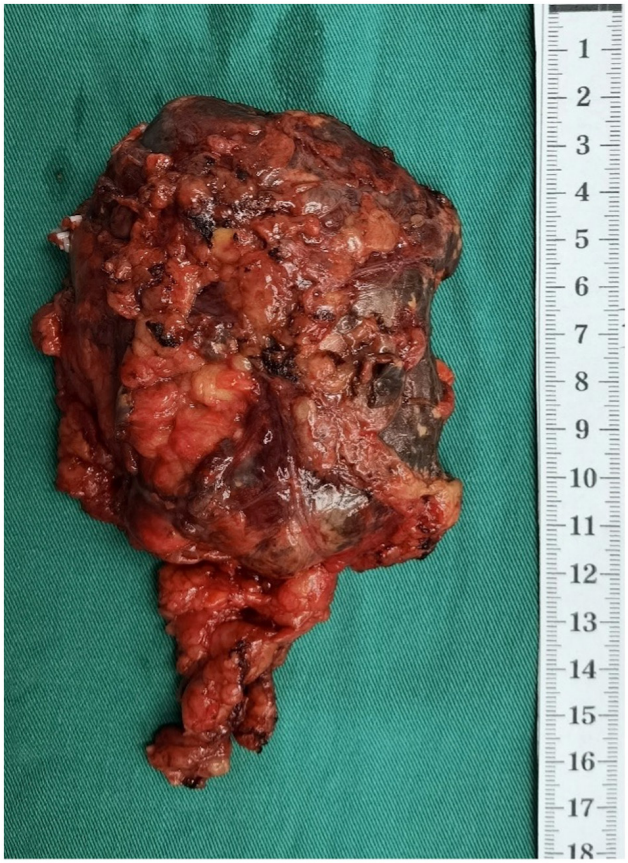

Figure 5. Macroscopic view of the resected giant right myelolipoma.

tumor rupture, bleeding, infection or tumor implantation. It is therefore not recommended for widespread use. Therefore, the gold standard for AML diagnosis remains postoperative histopathological examination.

Historically, AMLs larger than 6 cm in diameter were considered contraindications for laparoscopic surgery and managed with open adrenalectomy (25). However, with the advancement of endoscopic equipment and the refinement of laparoscopic and robotic surgical techniques, minimally invasive approaches have increasingly replaced open surgery even in large tumors (26-28).Growing experience and technological improvements have expanded the indications for laparoscopic management. Several studies have demonstrated the safety and feasibility of laparoscopic resection in giant AMLs (29). Laparoscopic adrenalectomy can be performed via transperitoneal or retroperitoneal approaches. The transperitoneal approach is often preferred for larger tumors requiring extensive exposure, particularly in obese patients or when tumors are located near critical structures such as the inferior vena cava, liver or other abdominal organs (30). This approach provides superior visualization and operative space, enabling safe, precise tumor resection while minimizing damage to vital vessels and organs, thereby reducing the risk of intraoperative and postoperative complications (31). By contrast, the retroperitoneal approach offers easier and direct access to the adrenal glands without transgressing the peritoneal cavity. This minimizes interference from intra-abdominal organs and facilitates quicker postoperative bowel function recovery and shorter hospital stays. Because it avoids major vascular

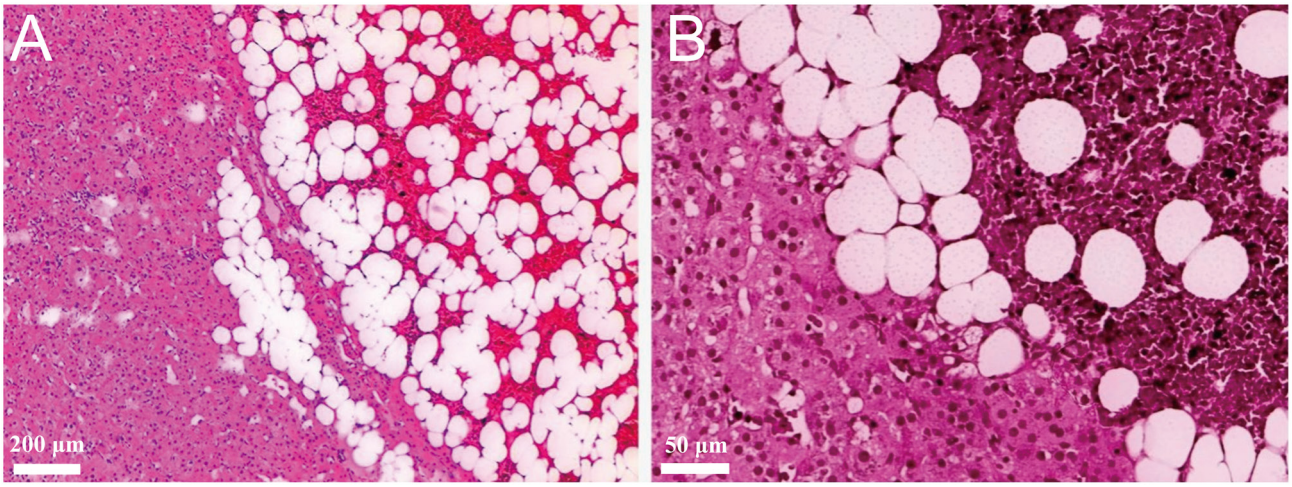

Figure 6. Photomicrographs of the resected myelolipoma (H&E staining). (A) Low-power view (magnification, 10x10; scale bar, 200 μm) with a well-circumscribed lesion composed of mature adipose tissue admixed with hematopoietic elements, alongside residual adrenal cortex. (B) High-power views (magnification, 10x40; scale bar, 50 μm), demonstrating an intimate admixture of adipocytes with trilineage hematopoietic cells

structures and organs, this approach is generally associated with reduced intraoperative bleeding, shorter operative time and lower rates of postoperative complications such as postoperative adhesions and infections (32). Our clinical experience also suggests that the retroperitoneal approach typically requires fewer trocars, further contributing to faster postoperative recovery. This approach is particularly suitable for relatively small tumors in favorable anatomical positions or in patients with a history of prior abdominal surgeries. A review of the literature shows that most unilateral AMLs are managed via laparoscopic adrenalectomy-predominantly through the transperitoneal approach, although the retroperitoneal route has also been successfully employed in selected cases (32). Bilateral AML is rare. When clinically appropriate, surgeons typically prioritize resection of the larger or symptomatic mass first, preserving the contralateral adrenal gland to maintain adrenal function, with periodic monitoring of the remaining lesion (33). The surgical strategy for unilateral resection in bilateral AML cases is similar to that for unilateral AML. However, when AML tumors are extensively adherent to major vessels or organs, when imaging or intraoperative findings raise suspicion of malignancy, or in cases of tumor rupture with acute hemorrhage, open surgery remains the recommended safer option for both unilateral and bilateral cases (14). Furthermore, in rare instances of bilateral giant AML where both tumors are large or high-risk, existing case reports predominantly describe bilateral adrenalectomy performed via transperitoneal laparoscopy or open surgery, with no cases performed via a retroperitoneal approach, and these patients require lifelong steroid replacement therapy postoperatively (34-36). No prior reports have documented bilateral adrenalectomy using both laparoscopic approaches (retroperitoneal and transperitoneal) in the same patient. Furthermore, comparative data between these two laparoscopic methods in the context of giant AML are currently lacking, to the best of our knowledge. In this case, after thorough preoperative evaluation, both retroperitoneal and transperitoneal laparoscopic approaches were employed in the same patient to resect bilateral giant AMLs. The perioperative data for both surgical approaches were also summarized in Table SI/Appendix. Consistent with existing literature, laparoscopic adrenalectomy was associated with several advantages, including reduced blood loss, less postoperative pain, low complication rates, faster recovery and shorter hospital stays (37,38). The present findings confirmed these benefits in both procedures. Based on perioperative comparisons and clinical experience at our hospital, it may be suggested that the retroperitoneal approach may offer certain advantages in selected cases. Although this report of a single case and lacks sufficient credibility with extensive data and a large sample size, it contributes valuable clinical insight into the surgical management of rare bilateral giant AMLs.

Furthermore, several factors may be considered to be crucial for achieving optimal outcomes in the laparoscopic resection of giant AMLs. First, during perioperative management, blood pressure, heart rate, serum potassium and adrenal hormone levels should be routinely monitored to detect and prevent potential functional lesions. Second, because AMLs often have a delicate capsule, dissection should be carried out carefully along the extracapsular plane to preserve its integrity, thereby reducing the risk of rupture that could obscure the operative field. Leaving a thin layer of pericapsular fatty connective tissue may further minimize the chance of capsule disruption caused by direct traction. Third, when the tumor is situated on the right side and lies close to the inferior vena cava, the surgeon must have a detailed understanding of the local anatomy and proceed with particular caution to avoid vascular injury. Finally, in bilateral AML resections, the risk of adrenal crisis is greater than in unilateral cases, making postoperative prophylactic hormone replacement therapy advisable.

The present case demonstrates that both retroperitoneal and transperitoneal laparoscopic approaches are safe and effective for the resection of giant AMLs. The retroperitoneal approach may offer advantages in selected cases, particularly when tumor location and size are favorable. Ultimately, the choice of surgical approach should be based on tumor size, location, relationship with surrounding organs, and the surgeon's experience and expertise.

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Availability of data and materials

The data generated in the present study are included in the figures and/or tables of this article.

Authors' contributions

XL and JY were the patient's attending physicians. JY performed the surgery. JW, WZ, YC and JL were involved in the collection of clinical and imaging data, preliminary analysis and figure preparation, and contributed to drafting the original version of the manuscript. YL was responsible for long-term patient follow-up and contributed to critical revision and editing of the manuscript. JW and JY confirm the authenticity of all the raw data. All authors have read and approved the final manuscript for submission.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

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