



Case report

Repair of multiple abdominopelvic aneurysms: is the open approach still valid? A case report

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ABSTRACT

Multiple Aneurysmal Arterial Disease (MAD) is an extremely rare arterial vascular condition and is produced by an abnormal alteration of smooth muscle cells and neutrophils, producing a multiple-aneurysmal degeneration. We present the case of a 36-year-old patient with a MAD in the cerebral territory and extremities with no surgical indication; however, with an aneurysm of the right inferior renal segmental artery, inferior mesenteric artery, left common iliac artery, and right internal iliac artery with surgical indication. An open approach with single-stage surgical repair, including graft interposition, bypass, exclusion, and vascular reimplantation, was performed. The surgical and postoperative course was uneventful, and the patient was discharged with an indication for outpatient follow-up. The open approach may be the best option for young patients with MAD, especially in the abdominopelvic region, without the need for high resources, with satisfactory results and improved patient survival.

Keywords: Aneurysm; Vascular Surgical Procedures; Vascular Graft (Source: MeSH-NLM).

RESUMEN

Reparación de aneurismas abdominopélvicos múltiples: ¿sigue siendo válido el enfoque abierto? Reporte de un caso

La enfermedad arterial multianeurismática (EAM) es una afección vascular arterial extremadamente rara y se produce por una alteración de las células musculares lisas y los neutrófilos, produciendo una degeneración multianeurismática. Presentamos el caso de un paciente de 36 años con una EAM en territorio cerebral y extremidades, sin indicación quirúrgica; sin embargo, con un aneurisma de la arteria segmentaria renal inferior derecha, arteria mesentérica inferior, arteria iliaca común izquierda y arteria iliaca interna derecha con indicación quirúrgica. Se realizó un abordaje abierto con reparación quirúrgica en un solo tiempo que incluyó interposición de injerto, bypass, exclusión y reimplante vascular. La evolución quirúrgica y posoperatoria transcurrió sin complicaciones y el paciente fue dado de alta con indicación de seguimiento ambulatorio. El abordaje abierto puede ser la mejor opción para pacientes jóvenes con EAM, especialmente en la región abdominopélvica, sin necesidad de altos recursos, con resultados satisfactorios y mejoría en la supervivencia del paciente.

Palabras clave: Aneurisma; Cirugía Vascular; Injerto Vascular (Fuente: DeCS-BIREME).

Introduction

Multiple Aneurysmal Disease (MAD) is an extremely rare arterial vascular condition (2-12%), with at least 200 cases reported worldwide.⁽¹⁻³⁾ This condition is caused by an abnormal alteration of smooth muscle cells and neutrophils, generating elevated intracellular elastase and leading to aneurysmal degeneration of the vascular wall at multiple locations in the body.⁽³⁾ It is associated with genetic or unknown conditions in 55% of cases, 15% connective tissue disorders, and 8% vasculitis.⁽²⁾ MAD often presents from early life to early adulthood and is mainly located in arteries of the extremities, aorta and great vessels, visceral vessels, and the brain.^(4,5) Diagnosis is often incidental or when there is a complication of the aneurysm; therefore, surgical treatment considers the independent criteria of aneurysm management according to the vascular sector affected, availability of materials, patient stability, and location. The open approach may be the best choice in young patients with MAD, especially of abdominopelvic location, and without the need for high resources, with satisfactory results and improved patient survival.^(6,7)

Case report

A 36-year-old woman with a congenital history of multiple small-caliber arterial aneurysms in both brachial arteries and the left anterior tibial artery. During childhood, an anatomopathological study was performed with a biopsy of the left radial artery, with no clear etiology identified but with a diffuse pattern of smooth cell dysplasia, concluding with the probable diagnosis of MAD. She did not present any intercurrent until the age of 32, when she presented symptoms of acute abdominal pain, identifying the rupture of a 5 cm fusiform left renal aneurysm; an extensive median laparotomy and emergency nephrectomy were performed.

On subsequent outpatient examinations, angiotomography identified in addition to the already known aneurysms, a cerebral aneurysm of the cavernous sinus (7 x 9 mm), Right Inferior Segmental Renal Artery Aneurysm (RISRAA) (3.5 x 4 cm), Inferior Mesenteric Artery Aneurysm (IMAA) (4 x 5 cm), Left Common Iliac Artery Aneurysm (LCIAA) (5 x 6 cm), Right Internal Iliac Artery Aneurysm (RIIAA) (3.5 x 4 cm), and without presence of coronary aneurysms (**Figure 1**). The multidisciplinary surgical assessment concluded a low risk of rupture of the cerebral aneurysm,

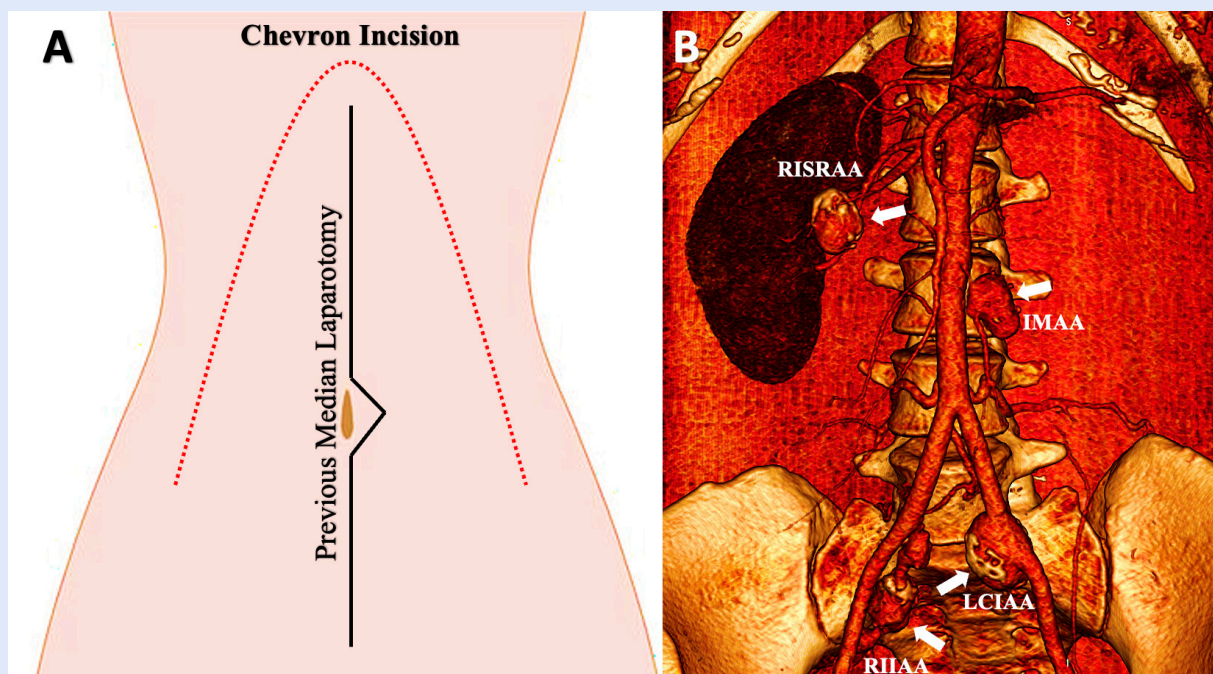


Figure 1. Open surgical approach. **A)** Chevron Incision. **B)** Tridimensional tomographic reconstruction of the multiple abdominopelvic aneurysms such as RISRAA, RIIAA, LCIAA, and IMAA, respectively, pointed out by white arrows.

RISRAA: Right Inferior Segmental Renal Artery Aneurysm. RIIAA: Right Internal Iliac Artery Aneurysm. LCIAA: Left Common Iliac Artery Aneurysm. IMAA: Inferior Mesenteric Artery Aneurysm.

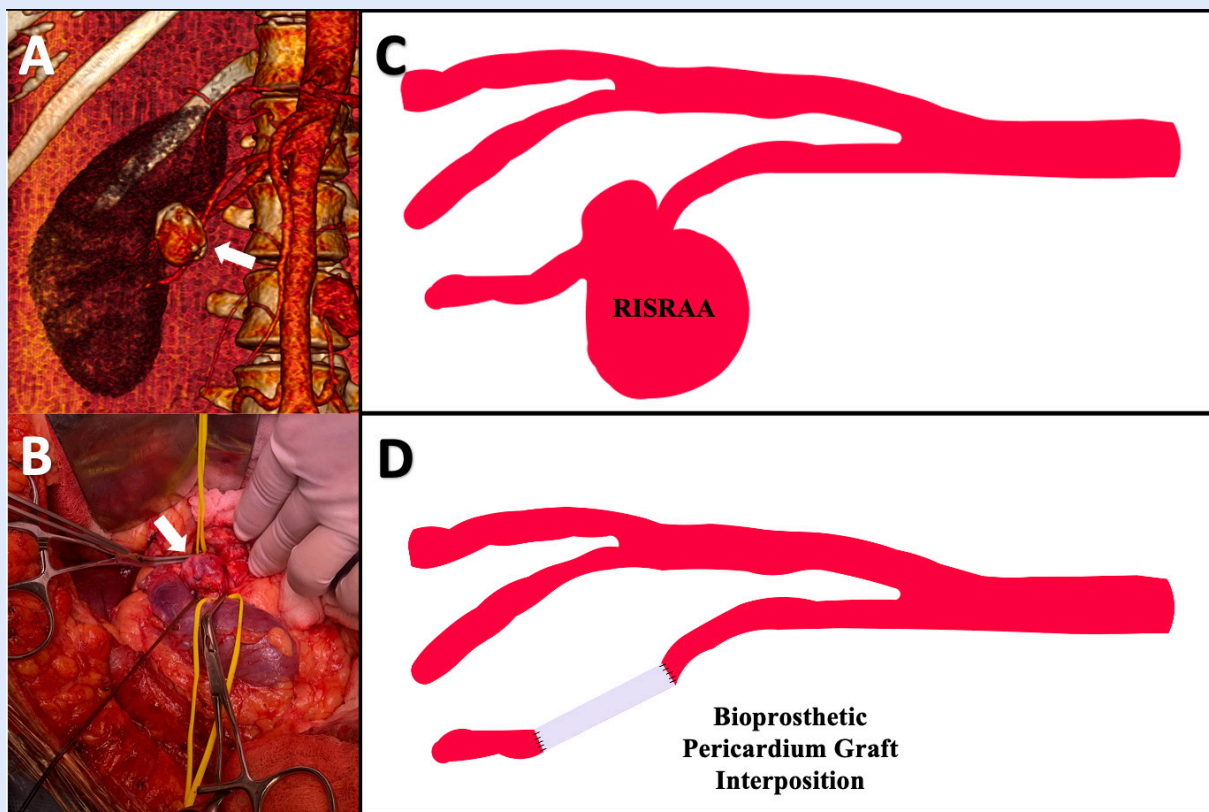


Figure 2. RISRAA repair. **A)** Tridimensional tomographic reconstruction of RISRAA showing a size of 3.5 x 4 cm (White Arrow). **B)** RISRAA open repair using a bioprosthetic pericardial graft interposition (White Arrow). **C-D)** Graphic description of RISRAA open repair.

RISRAA: Right Inferior Segmental Renal Artery Aneurysm.

indicating observation; however, for newly found aneurysms, it was necessary to consider various factors such as the risk of right nephrectomy and subsequent chronic kidney disease with hemodialytic support. On the other hand, the history of previous abdominal surgery was a latent challenge to consider for a future open approach; however, endovascular treatment was ruled out due to the risk of distal renal embolization, a complex anatomical configuration of the rest of the aneurysms and lack of supplies.

An open approach using an extended chevron incision with safe release of adhesions was indicated (Figure 1). For the RISRAA, a bioprosthetic pericardial graft interposition (diameter 6 mm) with selective clamping was performed. The IMAA was disinserted from the aorta with a proximal 4/0 polypropylene loop, and exclusion of the aneurysmal sac was performed (Figure 2). The distal, healthy portion of the Inferior Mesenteric Artery (IMA) was made ready for subsequent reimplantation. For the LCIAA, a Dacron prosthetic graft bypass was performed from the proximal region of the common iliac artery to the external iliac artery, excluding the aneurysmal sac next to the left internal iliac artery because of its proximity; also, the IMA was reimplanted over this Dacron graft with a lateral-lateral anastomosis (Figure 3). The RIIAA was ligated and excluded from the iliac blood flow

(Figure 4). The surgical procedure was performed without any intercurrent and during the postoperative period, the patient did not present any complications.

The anatomopathological study did not differ from that previously described and confirmed the diagnosis of MAD based on the Microscopic examination of the surgical pieces; therefore, it demonstrated intimal and medial fibrosis, along with fragmentation and a loss of elastin and smooth muscle fibers. Notable variability in smooth muscle cell proliferation was observed, accompanied by a discernible increase in collagen or ground substance within both the intima and media. No significant evidence of inflammatory cell infiltration was present. The patient was discharged from the hospital 5 days after the procedure with an indication for outpatient follow-up.

Discussion

Multiple Aneurysmal Disease (MAD) remains an unusual vascular pathology, with no previous incidence studies and fewer cases reported worldwide to date.⁽³⁾ Some authors have also

described it as a multi-aneurysmal, pan-aneurysmal, or polyfocal aneurysmal disease.⁽⁶⁾ The cause of this pathology is submerged in a wide range of diagnostic possibilities, such as connective tissue disorders (Ehlers-Danlos syndrome and Marfan syndrome), different types of arteritis and the wide clinical field of vasculitis, such as Wegener's granulomatosis, Takayasu's disease, Kawasaki's disease, and fibromuscular dysplasia, among others; taking into account that many of the published case reports presented an undefined diagnosis.^(2,9)

The age of presentation is variable, with no defined age groups pattern due to the low number of reported cases and due to the often-incident diagnosis or sudden diagnosis when it occurs with complications like rupture. Morphologically, saccular aneurysms are more frequent and have a higher rupture rate. Although MAD can have a mortality rate of 25-50% in cases of complications such as rupture of one of the aneurysms, no standardized and agreed treatment has been established.^(4,9) Management options include open surgery, endovascular approach with stent placement, embolization of lesions, and the indication for surgical procedures depending on the individual characteristics of the aneurysm (size, growth rate, morphology, neck) and the risk/benefit to the patient.⁽⁵⁾

Our case was evaluated by a multidisciplinary team headed by the Vascular and Endovascular Surgery Unit because it was necessary to address abdominopelvic aneurysms that

compromised a valuable kidney (mono-renal patient), the irrigation of the gastrointestinal system, and the lower limbs. Some reports recommend open surgery with the use of autologous and/or prosthetic grafts due to its viability and excellent results, especially in young patients, and with the benefits of being able to address all the abdominopelvic lesions that arise in the same surgical act with the acute complications that may occur, in addition to offering greater durability over time and less recurrence in the follow-up.^(6,8,10)

In our case, surgical management was chosen due to the size of the aneurysms, the history of emergency left nephrectomy for aneurysmal rupture, and the risk of contralateral aneurysmal rupture and, after evaluating the anatomy of the lesions, it was decided that an open approach was the most appropriate in this particular case because most of the aneurysms did not have an adequate neck, which is essential for placement of a stent device, and also there was a shortage of supplies for endovascular management. Also, with the endovascular approach, there was a high risk of migration of coils or embolizing substances in the renal segmental artery, which could cause partial or total infarction of the only kidney in a young patient, leading to lifelong dialysis renal replacement therapy.⁽³⁾ On the other hand, cases with successful treatment by total endovascular surgery or hybrid surgery (open and endovascular) have been reported.^(4,8) However, it is worth mentioning that these cases had minimal

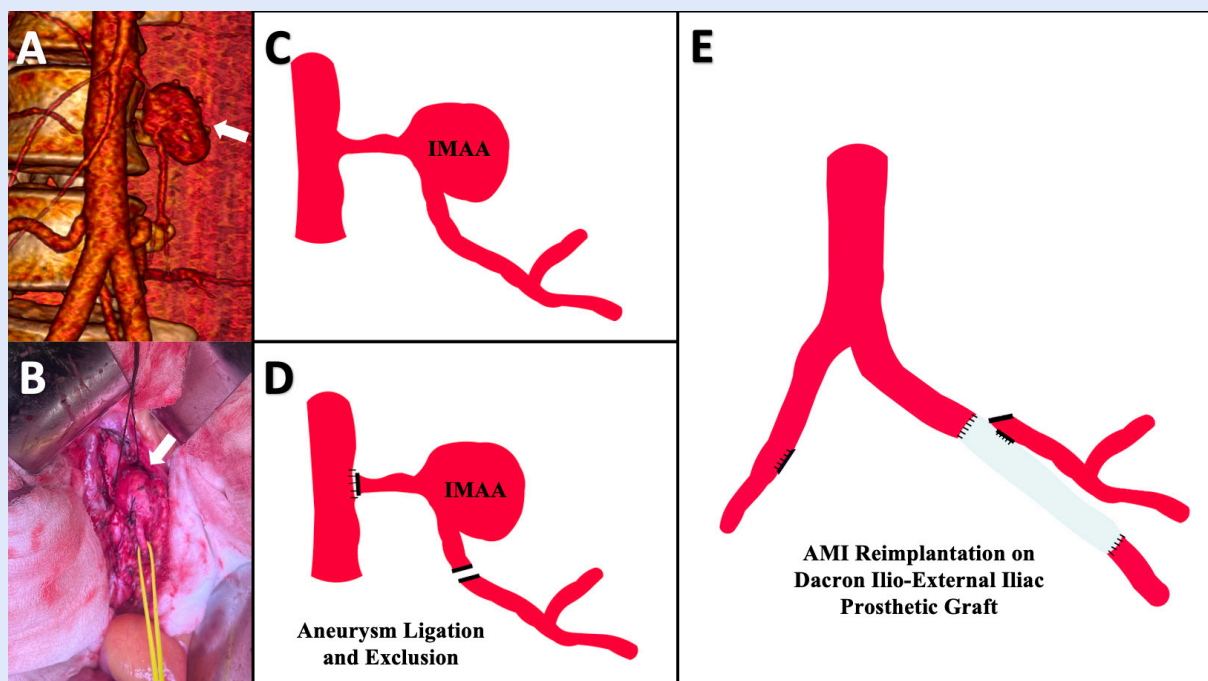


Figure 3. IMAA repair. **A)** Tridimensional tomographic reconstruction of IMAA with a size of 4 x 5 cm (White Arrow). **B)** IMAA open repair with surgical exclusion and next reimplantation to graft LCIAA (White Arrow). **C,D,E)** Graphic description of IMAA open repair.

IMAA: Inferior Mesenteric Artery Aneurysm. LCIAA: Left Common Iliac Artery Aneurysm.

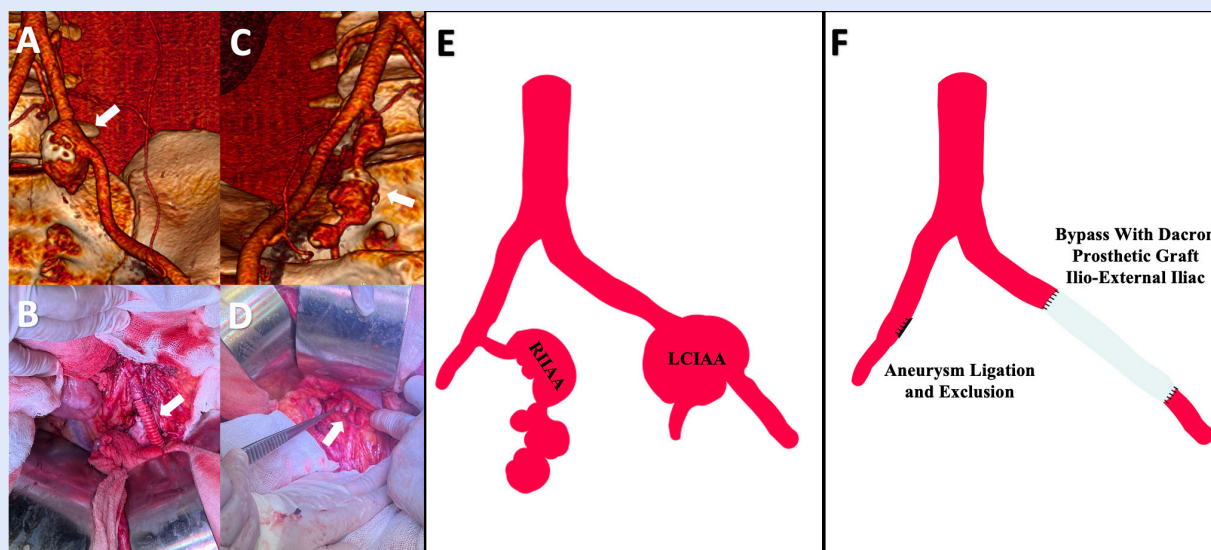


Figure 4. RIIAA and LCIAA repair. **A-C)** Tridimensional tomographic reconstruction of RIIAA and LCIAA with sizes of 3.5 x 4 cm and 5 x 6 cm, respectively (White Arrows). **B-D)** RIIAA and LCIAA are open repairs based on surgical exclusion and dacron prosthetic graft bypass, respectively (White Arrows). **E-F)** Graphic description of RIIAA and LCIAA open repair.

RIIAA: Right Internal Iliac Artery Aneurysm. LCIAA: Left Common Iliac Artery Aneurysm.

favorable aneurysmal morphology for such intervention and success rates around 80%.^(7,10) In addition, they did not present high risks of organ loss compared to our patient, reinforcing the open surgical approach as the most appropriate for this case.

Some authors have also described cases with expectant management in small aneurysmal lesions that did not irrigate vital organs and had a low growth rate and minimal risk of rupture.^(6,11) It has been found in case series that, during expectant management follow-up, visceral aneurysms had a spontaneous rupture rate of 10-20% and a mortality that can reach 50% in case of inadequate follow-up or undiagnosed patients debuting with ruptured aneurysm.^(4,9)

The diagnostic and planning study is performed by Angio tomography in most cases as the test of choice, in addition to the use of other studies that have been used in the reports, such as Doppler ultrasound as an initial study in limb lesions, angiography, and magnetic resonance imaging, which are used according to the location of aneurysms and comorbidities of each patient. It is recommended that when an aneurysm is found, the presence of other associated lesions should be ruled out with the aforementioned imaging methods, and they are also useful

for proper follow-up in patients under observation and in the postoperative period of patients undergoing surgery.^(5,8)

In conclusion, with the results obtained, without complications, with a short hospital stay after open surgery of a considerable magnitude, we believe that the surgical option used was the best for this case, managing to preserve the patient's only kidney without renal injury, without affecting urinary flow or renal filtration, without compromising the gastrointestinal circulation or the lower limbs. Ideally, there should be standardized management agreement in management guidelines; however, the scarcity of reports of this infrequent pathology still presents an obstacle to this end.

Authors' contributions

WSC, FAS, MSC, JMR, CPC, HL, ASH, JPS, NMN: Conception and design of the study. **WSC, FAS, MSC, JMR, CPC, HL, ASH, JPS, NMN:** Data acquisition, analysis, and interpretation. **WSC, FAS, MSC, JMR, CPC, HL, ASH, JPS, NMN:** Article writing, and critical review of intellectual content. **WSC, FAS, MSC, JMR, CPC, HL, ASH, JPS, NMN:** Final approval of the version to be published.

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