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Successful management of abdominal wall arteriovenous malformation by embolization and surgery

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Abstract:

Background: Congenital vascular malformations CVMs are usually challenging for the physicians, which need a multispecialty management, approach. Among these malformations, arteriovenous malformations are of special importance because of their ability to increase in size and bleed, and the hymodynamic effects of the areteriovenous shunting. Additionally, they never regress spontaneously.
Presentation of the case: 20 years old patient presented to the clinic with a congenital 20*40 cm painful hump on the lower left quadrant of the abdomen with pain, bleeding and ulceration. The patient was treated by endovascular emboliztion of the AVM feeding vessels, followed by surgical exicion and a plastic surgery to cover the resulting defect in the abdominal wall
Results: the resection was curative without major complications and the scar was very good 3 months after treatment without signs of relapse.
Conclusion: further periodic follow-up is required because of its high relapce ability taking into consideration the female gender of the patient.

Key words: Arteriovenous malformations, Embolisation, Vascular surgery, plastic surgery.

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التدبير الناجح لتشوه شرياني وريدي في جدار البطن بالتصميم والجراحة

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الملخص:

خلفية البحث وهدفه: تمثل التشوهات الوعائية تحدياً للأطباء المعالجين، عادة ما يحتاج تدبيرها إلى فريق طبي متعدد الاختصاصات، من بين التشوهات الوعائية تمثل التشوهات الشريانية الوريدية أهمية خاصة بسبب قابليتها للزيادة بالحجم والنزف وإحداث قصور قلب عالي النتاج، كما أنها لا تتراجع عفويا،

مواد البحث وطرائقه: تم إجراء البحث في مشافي جامعة دمشق، حيث راجعت المريضة ب

شكاية بروز مؤلم في الخاصرة اليسرى، مع قصة تقرح ونزف والم وزيادة في الحجم خلال السنتين الأخيرتين، تم بداية تصميم الأوعية المغذية للتشوه الوعائي، ثم بعد شهر إجراء استئصال الآفة جراحيا، و تم بزمن ثالث تغطية الضياع المادي بواسطة شريحة إليوية ذاتية.

النتائج: تم تدبير المريضة بشكل ناجح، باستئصال التشوه الشرياني الوريدي بشكل جيد، مع تجنب حصول نزف كتلي حول الجراحة، وغياب علامات للنكس، وتغطية الضياع المادي التالي لاستئصال الورم مع تندب جيد

الاستنتاج: ، مع التأكيد على ضرورة متابعة المريضة بشكل دوري لتحري علامات النكس.

الكلمات المفتاحية: جراحة الأوعية الدموية، الجراحة التجميلية، شريحة إليوية، التصميم عبر اللمعة، التشوهات الشريانية الوريدية

1. Introduction:

Vascular anomalies is a term used to describe two divert groups of vascular lesion: vascular tumors and vascular malformations (Lee BB *et al*, 2015, 333 – 374) [(1)].

Vascular tumors are lesions which result from true proliferation of endothelial cells, while congenital vascular malformations describes the malformed vessels which result from arrested development of the vascular system at different phases of embryogenesis (Enjorlas *et al*, 1997, 375-423)[(2)]

The divert histological sources and the different developemental stages at which the developmental arrest happens leads to wide range of clinical presentations, unpredictable clinical courses, and erratic responses to treatment, along with potentially high rates of recurrence (Lee BB, 2002, 209 – 213).[(3)]

2. Classification:

The nomenclature and classification of congenital vascular malformation had been confusing, regarding its various types. The most common classifications of these lesions is the Hamburg classification, which classifies these lesions depending on the predominant histological component into: venous malformations, arterial, arteriovenous, mixed hemolymphatic and capillary malformation.

Each one of these histologic subtypes is further classified into extra-truncural and truncural forms (belov, 1989, 25-30). [(4,5)]

Another important classification is the Mulliken/ISSV classification, which divides these malformations into 2 groups: fast flow lesions and low flow lesions. This classification is important because the flow velocity is important to predict the hemodynamic and clinical effects of each lesion (Mulliken JB *et al*, 1982, 412 – 420)[(6)]

Arteriovenous malformations (AVMs) are high flow malformation characterized by abnormal connections between feeding arteries and draining veins (Lee BB *et al*, 2006, 597 – 607) [(7)]

An avm may begin as only a pink stain, but can expand due to female hormonal changes like pregnancy and puberty, or secondary to local trauma. Expansion can lead to ischemia distal to the AVM, local hyperthermia, dialated viens, thrill and bruits. In addition to pain, ulceration, functional impairment, soft tissue and bony destruction, and bleeding (LeeBB *et al*, 2016, 9-36)[(8)] These symptoms necessitate treatment. Larg avms can also cause high-output cardiac failure, which requires early intervention (Lee BB, 2009, 193-200)[(9)] An asymptomatic lesion can be observes, although it is preferable to treat befor the AVM progresses (LeeBB *et al*, 2016, 9-36)[(8)]

3. Case report:

A 18-years-old female presented with a congenital (40 * 20 cm) lump in the left middle and lower quadrant of the abdominal wall. The lump appear after birth and was increasing in size for the past two years. She presented with painful and bleeding ulcer. Non-tender, non-pulsatile, no local hyperthermia

Clinical vascular examination reveal a thrill and wide pulse on left femoral artery and vein.

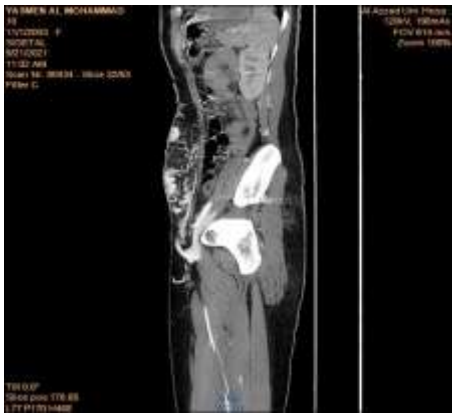
Laboratory tests;

Wbc	5.6/59.5	alb	4.2
Hb/ht	10.6/33.7	Esr	80
Plt	365	Crp	2.8
Cr	0.8	Blood group	B^+

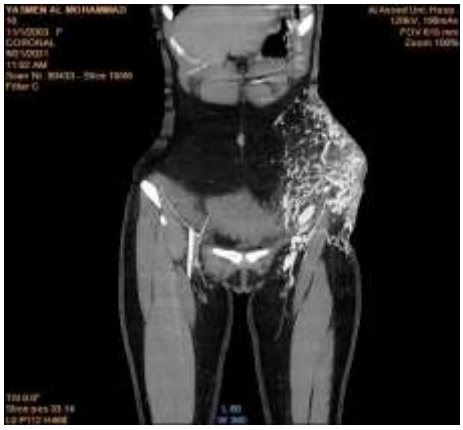
Fig (4): laboratory tests results of the case before surgery.

Ultrasound; large hyperechoic mass in the left middle and lower abdominal wall expanding to deep soft tissue involving a large number of tortuously, dilated vascular vessels measured about 2-14 mm, without thrombosis. Suggesting capillary hemangioma.

MSCT showed Excessive abnormal subcutaneous perfusion at the left iliac flank, extending posteriorly. Perfused from the intercostal arteries 11-12, also from left internal iliac artery and the common femoral artery.



a



b

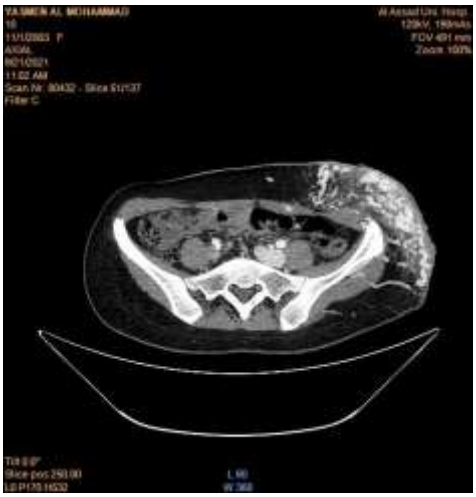
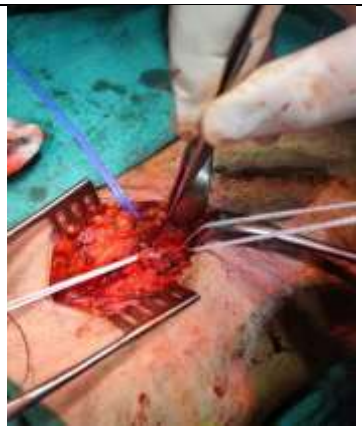


Fig (1): MSCT of the case lesion: a: para-median saggital section, b: frontal section c:cross section.

Surgery (23/9/2021); the patient underwent an embolization session through a contralateral approach of large gluteal arteries by selective engagement of left internal iliac artery. After one month the patient underwent wide excision of the lesion after ligation of the tribulation arteries of the lesion. After resuscitations, the patient underwent a skin grafting reconstruction of the lesion supernal surface.

Pathology showed scattered arteries and veins with variable walls' thickness with channels connecting them without capillary beds and only small quantities of normal tissue were observed in the lesion.



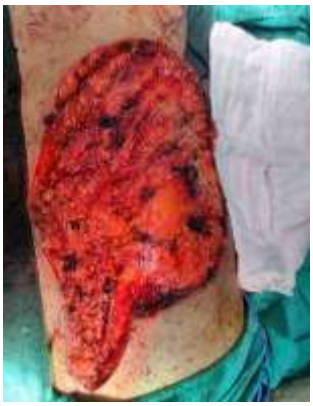


fig (2): after surgical excision of the lesion

Follow up: The patient was followed up on the first day in the intensive care, then in the vascular surgery department for two days, and then she graduated to the plastic surgery department at Al-Mowasat University Hospital in Damascus, where a skin graft was performed for the excision area and she graduated home after 7 days. The time between excision and skin grafting was 12 days. During this period, the patient was in good general condition and did not develop any complications (bleeding, fever, ...). An excellent wound healing result was reached after approximately 80 days.

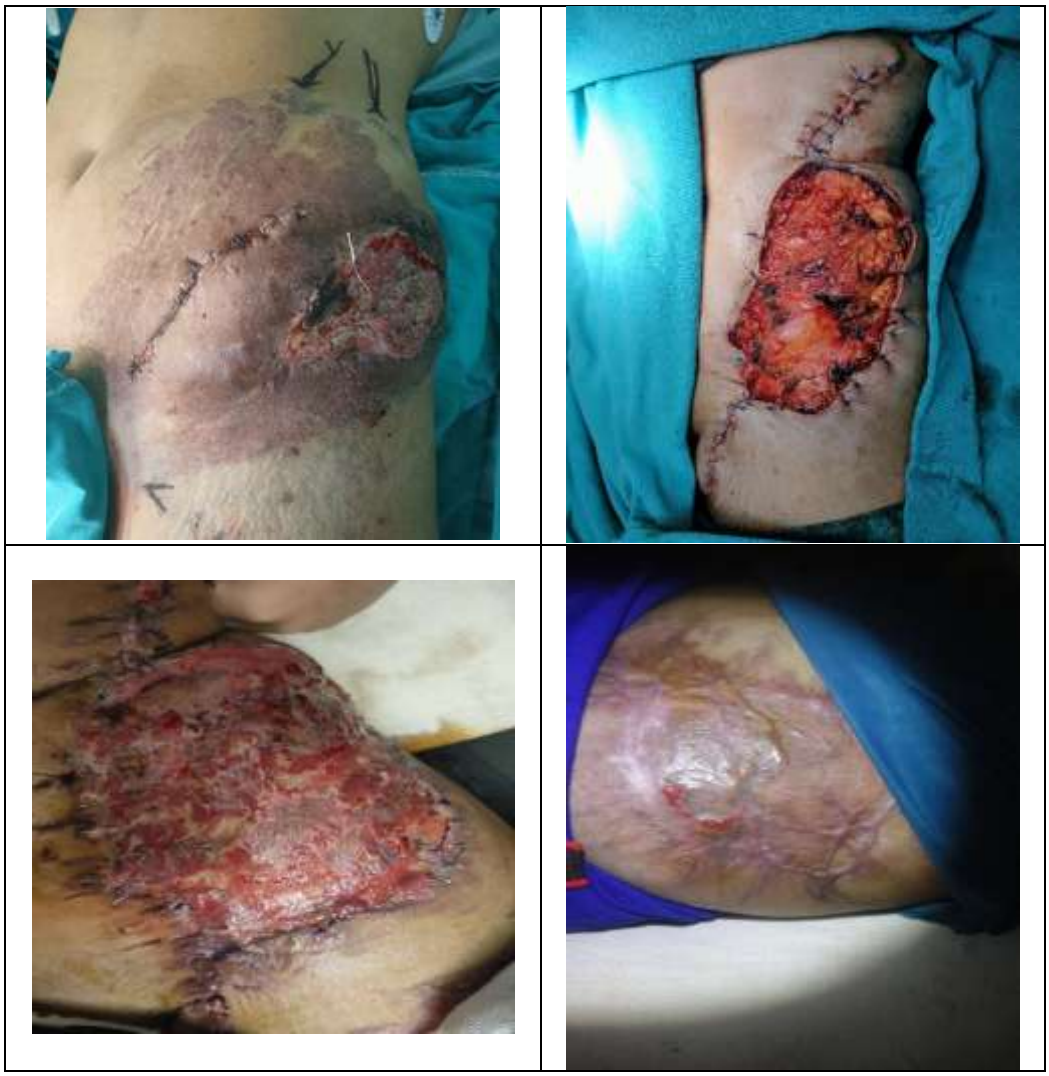


Fig (3): stages of the plastic management of the lesion.

Discussion:

Congenital Vascular Malformation (CVM) terminology is used to describe malformed vessels resulting from the arrested development during various stages of embryogenesis. According to Hamburg classification, AVMs represent one of the five classes of CVM and according to ISSVA classification, AVMs are categorized as fast-flow lesions.[6] A systemic review of literature found that the incidence of cerebral AVMs, the commonest presentation of AVM, ranged from 1.12 to 1.42 cases per 100,000 every year with hemorrhage being the most common presentation.[7] The pathophysiology of AVMs is still unclear and might have a genetic basis. AVMs do not usually become apparent until the first or second decade of life and can dangerously grow rapidly. Symptoms vary depending on the size and place of the lesion as they can be asymptomatic but can become lethal when complicated. Trauma, hormonal changes during puberty and pregnancy, biopsy and inappropriate treatments are considered as stimulation factors for the progression of AVMs. Diagnosis can be misleading and challenging; Doppler ultrasonography can demonstrate shunts with high-flow lesion while MRI can confirm the diagnosis. The gold standard imaging modality is DSA which is also indicated with intent-to-treat by embolization or resection. Treatment options include embolization, surgical resection or a combination and currently there is no pharmacologic treatment. Pre-operative embolization is needed in large lesions in order to reduce blood loss. Excisional surgery should be implemented from 24 to 72 h after embolization before recanalization and angiogenesis take place.[8] so that the efficacy and outcomes of the embolization are maximized. In conclusion, any cutaneous pigmentation should be evaluated carefully in order to have a correct diagnosis to distinguish between a benign lesion and congenital vascular malformation (CVM). In case of CVM, earlier treatment leads to better results and can help preventing such a trauma and low quality of life for a child in such a young age and financial hurdles should be overcome as this is a priority for the child.[9]

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