CASE REPORT

A Unique Case: Arteriovenous Malformation of the Urinary Bladder Wall

S Cansaran[1], NI Atay[1], AK Pelin[1], AC Celayir[1], S Moralioğlu[1], N Koc[2]

ABSTRACT

Arteriovenous malformation (AVM) of urinary bladder is a very rare condition in which a section of blood vessels lacks capillary vessels resulting in blood from an artery being delivered directly to a vein. We report a rare case of AVM of the bladder wall mimicking a bladder tumour presenting with acute abdomen.

Keywords: Arteriovenous malformation, bladder, child.

INTRODUCTION

Arteriovenous malformation (AVM) of urinary bladder is a very rare condition in which a section of blood vessels lacks capillary vessels resulting in blood from an artery being delivered directly to a vein. The greatest danger is haemorrhage. Treatment for AVMs includes surgery or focused radiation therapy (1–3). We present a girl who had a mass with atypical localization arising from the bladder wall, diagnosed as arteriovenous malformation pathologically, due to very rare localization of AVM.

CASE REPORT

A 6-year-old girl was admitted to our department with sudden onset of abdominal pain, vomiting, dysuria and constipation. The past history revealed that she had been treated in another hospital due to severe constipation and haemorrhoids and rectal bleeding. Physical examination revealed acute abdomen, rebound tenderness, suprapubic abdominal mass and severe haemorrhoids. Pelvic ultrasound showed a central hypoechoic lesion sized 64 × 43 mm, surrounded by round hyperechoic area located from suprapubic areas to the Pouch of Douglas, suggesting the existence of appendicular abscess. A suprapubic mass was palpated under emergency general anaesthesia and a mid-line incision was made in the inferior abdomen. A 15-cm diameter, encapsulated mass arising from the anterior bladder wall was identified (Fig. 1). There were necrosis and haemorrhage in the centre of the mass. The mass was excised in its entirety from the bladder wall and inter muscular area. The inner surface of the bladder was clean, and there was no macroscopic anomaly. The huge arteriovenous malformation demonstrated central necrosis and inflammatory changes were identified by in pathological examination.

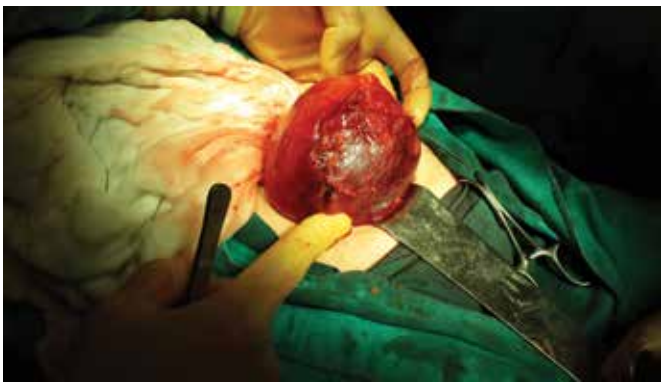

Fig. 1: A 15-cm diameter encapsulated mass arising from anterior bladder determined during the laparotomy under the emergency circumstances.

Section of the lesion had dirty yellow colour, and haemorrhagic and necrotic portions (Fig. 2). Oedematous stroma which is rich with chronic inflammatory cells and fibroblasts, enlargement of vessels lined by single layer of endothelium and irregular vascular spaces were observed microscopically (Fig. 3).

Post-operative course was uneventful, except for intravesical haemorrhage. Haematuria and haemorrhoids

From: [1]Department of Paediatric Surgery, Zeynep Kamil Maternity and Children's Training and Research Hospital, Istanbul, Turkey and [2]Department of Pathology, Zeynep Kamil Maternity and Children's Training and Research Hospital, Istanbul, Turkey.

Correspondence: Dr S Moralioğlu, Department of Paediatric Surgery, Zeynep Kamil Maternity and Children's Training and Research Hospital, 34668 Üsküdar Istanbul, Turkey. Email: serdarmoralioglu@gmail.com

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resolved 3 months later. There was only oedematous bladder mucosa in the cystoscopic examination 6 months after initial surgery. Her follow-up continues without any symptoms.

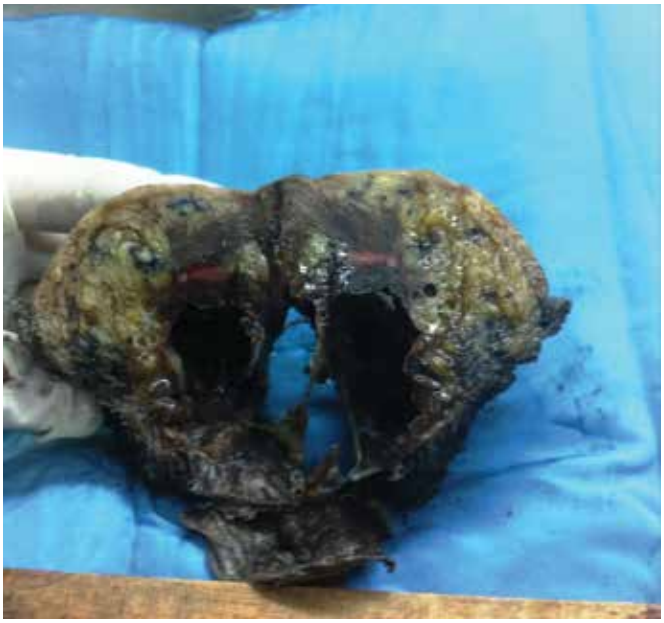

Fig. 2: The huge mass had demonstrated central necrosis, and inflammatory changes were identified macroscopically.

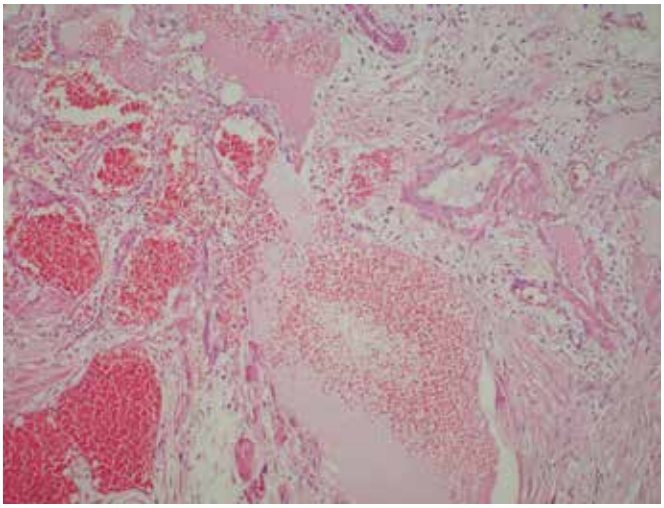

Fig. 3: Irregular vascular spaces in oedematous and inflammatory stroma were seen histopathologically (HE ×200).

DISCUSSION

Arteriovenous malformations are defects in vascular system. An AVM is a snarled tangle of arteries and veins. They are connected to each other, with no capillaries, which interfere with the blood circulation in an organ. The cause is not known, but they seem to develop during pregnancy or soon after birth (1–3).

Arteriovenous malformations most likely develop during the late somite stage in the 4th to 8th week of embryonic life. Early in the 3rd week of embryonic life, angioblasts begin to differentiate from the mesoderm, forming syncytial islands. These small clumps of syncytial cells develop tiny sprouts that interconnect the cell groups, forming a syncytial plexus that later differentiates into the primordial vascular plexus. These plexuses contain afferent, efferent and capillary portions. Arteriovenous malformations arise from persistent direct connection between the future arterial and venous sides of this primitive vascular plexus without the capillary component (1–4).

Arteriovenous malformations can be either congenital or acquired. It can occur anywhere, but they are more common in the brain or spinal cord. However, an AVM of the urinary bladder is extremely rare. They give different symptoms and signs depending on localization. AVMs occur in females more frequently than males (3). The index case was a 6-year-old girl with the congenital AVM on the bladder.

Therapeutic strategies change according to the size and the location of the AVMs (4–6). The optimal treatment has not been determined, but it is accepted that complete ligation of the afferent vessels and excision of the entire mass are essential for successful surgical treatment. In the symptomatic or rapidly enlarging lesions, especially for intramedullary AVMs, surgical treatment alone carries greater risks. Proximal ligation of the arterial supply of spinal cord AVMs could produce good short-term outcomes. On the other side, proximal ligation of the arterial supply of AVMs can lead to the formation of multiple small collaterals to the AVMs. It makes further embolotherapy more difficult. Bao et al. argued that, most intramedullary AVMs are inoperable, because the tangle of abnormal vessels fills almost an entire segment of the spinal canal and contains cord tissue within the interstices of the AVMs (5). With improved microsurgical technology, many intramedullary AVMs can be removed by using the same standard techniques used to treat intramedullary tumours.

Since the 1960s, endovascular therapy has been performed as an alternative treatment of AVMs. Preoperative particulate embolization markedly decreases intraoperative bleeding and makes the dissection of the nidus much easier (5). It also diminishes the threat of further haemorrhage and improves the perfusion of the spinal cord by diminishing the steal or venous congestion secondary to the malformation (6).

We used surgical therapy for this patient. The intermingling of the malformation required the surgical removal of lesion by total excision extravesically. The procedure resulted in remarkable improvement of the patient's symptoms, which had included gait disturbance provoked by pain in the lower extremities and haemorrhoids. The resolution of symptoms perhaps resulted from removal of the mass, which caused pressing of the pelvic and para rectal vessels, as well as from removal of the AVM, which relieved venous hypertension and congestion of the vessels of the rectum.

CONCLUSION

Arteriovenous malformations are extremely rare lesions of the urinary bladder. They mostly follow a clinically benign course but they are commonly misdiagnosed macroscopically as malignant tumour because of that their solid nature and ill-defined looking due to ulceration and necrosis of mass. We recommend careful pathologic assessment for establishing the appropriate diagnosis and either a conservative or aggressive surgical treatment for benign or localized mass of AVMs of the urinary bladder, respectively.

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