BRACHIAL ARTERY ANEURYSM IN A NEONATE

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Presented in the article is a clinical case report concerning successful surgical treatment of a rare disease – a progressing congenital true brachial artery aneurysm in a newborn girl. The symptoms were first noted at the age of 7 days, later on followed by enlargement of the aneurysm, appearance of neurological symptoms, and impaired function of the extremity. The infant was subjected to clinical examination and ultrasonographic study, followed by surgical removal of the aneurysm and restoration of the brachial artery by an «end-to-end» anastomosis. The diagnosis was finally verified only at histological examination. Also contained in the article is a review of the literature underlining that paediatric arterial aneurysms are extremely uncommon, as well as discussing the problems of diagnosis and therapeutic policy. The dilatation of the vessel turned out to be a true aneurysm with all three layers of the vessel in the wall and belonged to the class of paediatric congenital idiopathic arterial aneurysms unassociated with degeneration of the vascular wall or cardiovascular pathology.

Key words: *aneurysm, neonate, infant, diagnosis, treatment.*

INTRODUCTION

Congenital arterial aneurysms in children are extremely uncommon. This pathology is little-known and may conventionally be referred to rarely encountered diseases. Only sporadic case reports have been described in the world literature. Davis F., et al. (2016) reported the data of the University of Michigan Medical Center, describing 7 cases of aneurysms of arteries of upper extremities (of these, 3 cases concerning the brachial artery), with the youngest infant aged 4 months old. The same authors presented the data concerning a case report of treating a radial artery aneurysm in a two-month-old infant [1]. Sarkar R., et al. reported four cases of upper-limb aneurysms (with one of them being the brachial artery in a two-week-old infant) [2]. Other authors report only from 11 to 14 various clinical cases in the world literature in children less than 12 years old [3–5]. In the Russian-language literature no similar cases have been reported.

The development of paediatric aneurysms may be associated with such accompanying processes as infections, injury, connective-tissue diseases, arteritis or congenital vascular malformations [3, 1]. Only about 5% of arterial aneurysms in infants are located in the upper extremity and the majority of them are typically associated with systemic diseases [4].

Given exceptional rarity of the pathology, we present a clinical case report concerning diagnosis and successful surgical treatment of a true congenital progressing brachial artery aneurysm in a newborn infant.

Case report

Our patient was a girl born prematurely of the first pregnancy, with the gestation period being free from deviations. At the age of 7 days, her parents paid attention to a swollen mass in her left upper extremity in the area of the bend of the elbow. Family history: neither vascular malformations nor arterial pathology in the family were revealed.

The girl was subjected to ultrasonographic examination of soft tissues (age 7 days), revealing a rounded swollen mass in the lower third of the left shoulder, measuring 1 cm in diameter and 1.5 cm^3 in volume, with clear-cut uneven contours and the brachial artery pushed anteriorly. Emergency pathology was excluded. At that moment no data on the vascular origin of the morbid growth were obtained. A diagnosis of «hygroma» was suggested. Outpatient follow-up of the girl was initiated at the place of residence.

One month later, at the age of five weeks she was subjected to repeat examination showing growth of the swelling, with the appearance of pulsation felt on palpation. She was consulted by a paediatric vascular surgeon and diagnosed as having a rounded, elastic, clear-contoured pulsating mass in the area of the lower third of the left shoulder and bend of the arm, with additionally revealed oedema of the forearm and the hand on the left, decreased motion activity of the limb, with limited thumb and index finger flexion and the pulse on the arteries of the left forearm weakened (Fig. 1).

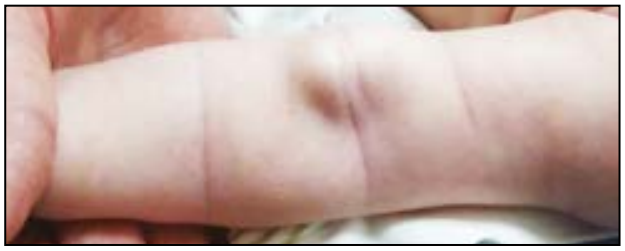

Fig. 1. Appearance of the left limb at admission to hospital, showing defiguration of the brachial area in the projection of the aneurysm

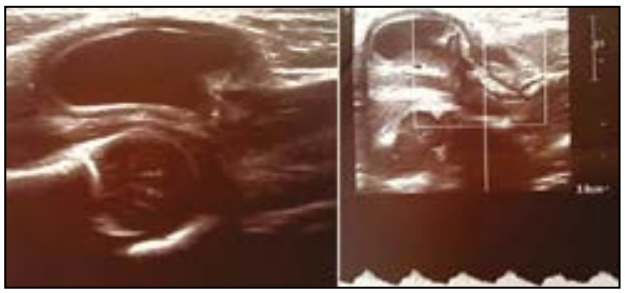

Fig. 2. Ultrasonographic examination of the brachial artery prior to surgery

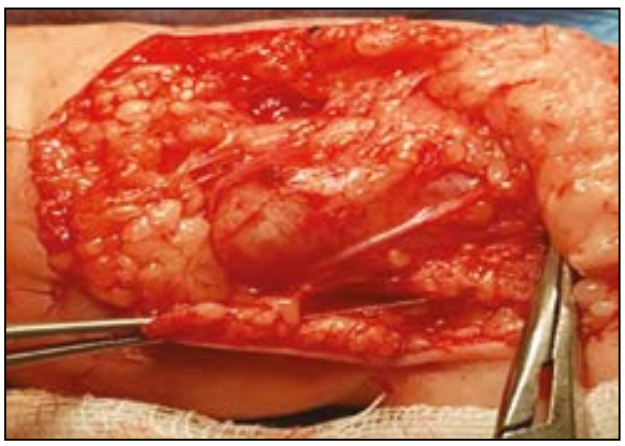

Fig. 3. Appearance of the brachial artery aneurysm after exposure

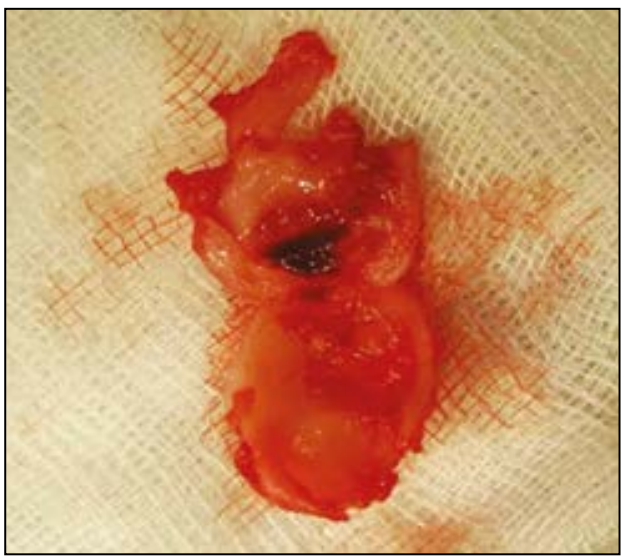

Fig. 4. Brachial artery aneurysm on the left after excision

Ultrasonographic examination was performed in the mode of colour Doppler mapping and energy Doppler. The diagnosis of a brachial artery aneurysm on the left was established. Examination revealed a 20x11 mm swollen mass, unechogenic, of irregular shape, with clear-cut smooth contours, with a 1.5-mm-thick capsule. The swelling was located along the posterior-medial wall of the brachial artery, pushing the artery upwards. Visualized were the proximal and distal portions of the left brachial artery, measuring slightly more than 1 mm in diameter. The size of the ostium between the artery and the aneurysmatic cavity was 1.1 mm, with the blood flow velocity in the ostium measuring 3.0 m/s. In the cavity of the aneurysm there was a low-speed turbulent blood flow. The brachial artery below the ostium of the aneurysmatic sac was flattened above the swelling and compressed. The blood flow through the artery was lowered. The blood flow distal to the aneurysm was with characteristics of collateral blood flow (Fig. 2).

Thus, a 2-fold increase in the size of the swelling was observed, accompanied and followed by evidence of limb ischaemia and functional impairments.

At the age of 2 months, the girl was hospitalized to the Department of Cardiovascular Surgery of the Republican Hospital, with the following diagnosis made: a congenital left brachial artery aneurysm. The infant was prepared for the operation.

At the age of 2 months she underwent reconstructive operation, under general anaesthesia, narcosis. An S-shaped incision in the left ulnar fossa was made. Exposed was a pulsating, round-shaped swelling measuring 23 mm in diameter and 35 mm in length. The brachial artery and the aneurysmatic sac were isolated (Fig. 3).

The aneurysm was incised (Fig. 4), with the length of the defect of the arterial wall after removal of the aneurysm and mobilization of the vessel's edges amounting to 15 mm. An «end-to-end» anastomosis of the brachial artery measuring 1.2 mm in diameter was applied, using thread 8/0. After the anastomosis was established the pulsation of the brachial artery was conducted along the whole length of the vessel. The technical possibility of forming the «end-to-end» anastomosis was achieved by performing postoperative immobilization in the position of the limb bent at 90°.

Histological examination of the removed material. A fragment of the vessel's wall with loosening of the tunica intima and tunica media, in the portions of endothelial desquamation there formed a thrombus with signs of organization. In the zone of the confluence into the aneurysm in the vicinity of the afferent artery there was cartilage-type thickening

of the tunica media, with the tunica media containing infiltration and vasculitis of small vessels. The walls of the aneurysm consisted of the same layers as the arterial wall, thus strongly suggesting the true nature of the aneurysm (Fig. 5).

The following diagnosis was confirmed: a true idiopathic (congenital) left brachial artery aneurysm.

The postoperative period. Anticoagulation therapy with unfractionated heparin was carried out for 13 days. Analgesics for 3 postoperative days. Vasodilators for 5 days postoperatively. From postoperative day 14, antiaggregant agents (acetylsalicylic acid) were prescribed for 3 months.

Ultrasonographic examination (14 days after the operation). The diameter of the brachial artery in the area of the anastomosis amounted to 0.8 mm, with the acceleration of blood flow through it up to 1.2–1.6 m/s and the blood flow through the brachial artery satisfactory, of the main type (Fig. 6).

The girl is on outpatient follow-up. The last examination was at 1 year of age, demonstrating that the left upper extremity was of physiological colour, warm, with the pulsation of the arteries of the forearm satisfactory. The findings of the ultrasonographic examination confirmed patency of the left brachial artery. The diameter of the artery in the area of the anastomosis increased to 1.8 mm. There were no neurological deviations (Fig. 7).

DISCUSSION

Congenital aneurysms of arteries of the upper extremities in children appear to be exceptionally uncommon. Only sporadic publications in the world literature have been devoted to their detection and treatment. A clinicopathological classification of arterial aneurysms in children was suggested by Sarkar R. (1991). According to this classification, aneurysms in children are subdivided into 9 classes. Class I – aneurysms associated with arterial infection, class II – giant-cell aortoarteritis, class III – autoimmune connective tissue disease, class IV – Kawasaki's disease, class V – Ehlers–Danlos syndrome or Marfan's syndrome, class VI – other forms of noninflammatory medial degeneration, class VII – arterial dysplasias, class VIII – congenital-idiopathic factors, as well as class IX – false aneurysms associated with extravascular events causing vessel wall injury or disruption [2]. According to the classification of Sarkar R. (1991), the case described herein belongs to class VIII, with the infant diagnosed as having an arterial aneurysm of congenital-idiopathic factors.

Currently, the cumulative number of the described case reports concerning patients less than 12 years old does not exceed two dozen, and the cases of successful detection and treatment of brachial artery aneurysms

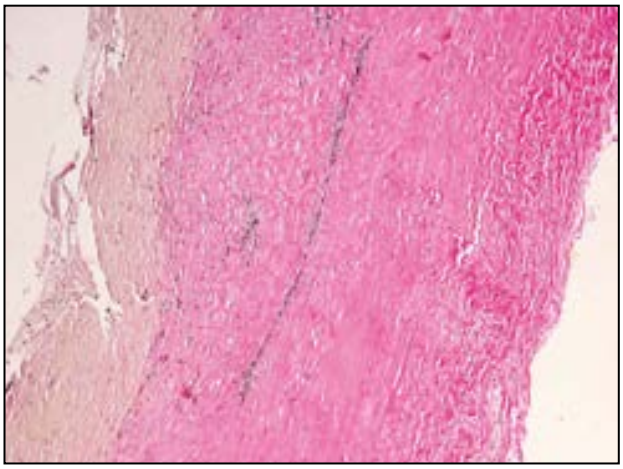

Fig. 5. Microscopic study of the aneurysmal wall. Haematoxylin and eosin stain, X 20. Microphotograph X 0.63

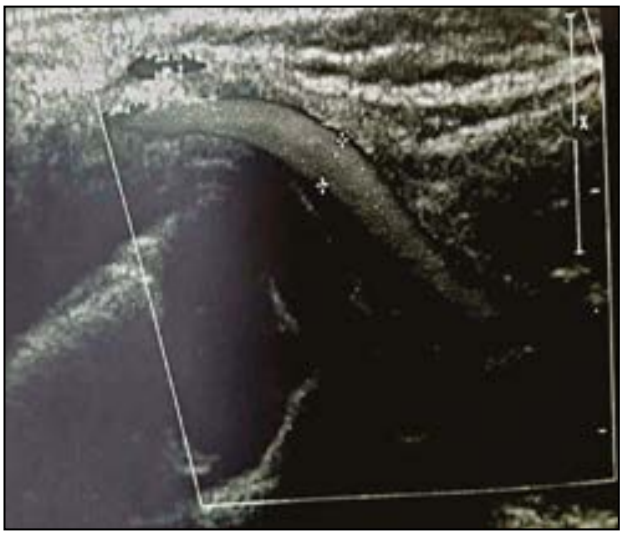

Fig. 6. Ultrasonographic examination. The brachial artery is patent

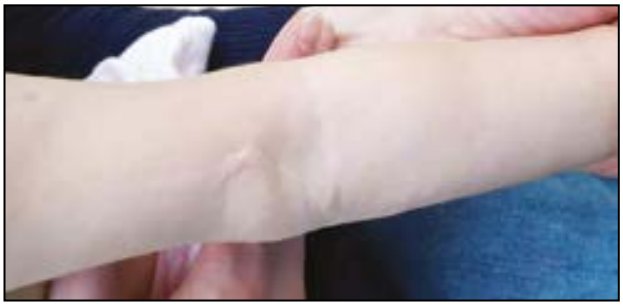

Fig. 7. Appearance of the limb one year after the operation

in newborn infants are unique. As a rule, paediatric aneurysms occur in the presence of one of background conditions.

Opinions concerning the terms of surgical treatment differ. Some authors consider that complications of "true" brachial artery aneurysms are uncommon and children who have them can be safely followed up for several years to permit growth of the artery before aneurysm resection. [3]. Other specialists suggest that

surgical intervention for upper extremity aneurysms should be initiated without delay in order to prevent extremity ischemia and amputation [6]. In the described clinical case there was progression of the disease with the appearance of and an increase in neurological symptomatology due to nerve compression by the aneurysm, venous insufficiency of the limb in the form of oedema, ischaemia and dysfunction of the extremity.

The experience of surgical treatment described in the article is characterised by several peculiarities. The diagnostic search was distinguished by uncertainty as to the vascular genesis of the detected swelling at the early stage of the disease, thus creating difficulties for early making an accurate diagnosis.

At the same time the aneurysm was a true aneurysm with all three layers of the vessel in the wall and belonged to the class of paediatric congenital-idiopathic arterial aneurysms unassociated with degeneration of the vascular wall or cardiovascular pathology.

In the described case we managed to successfully perform resection with an anastomosis, having avoided the need for prosthetic reconstruction of the artery. The intervention required a microsurgical technique of handling the vessel; to perform an «end-to-end» anastomosis required immobilization of the limb in the forced position.

CONCLUSION

Despite rarity of the pathology, timely and adequate surgical treatment of aneurysms of arteries of limbs in infants requires caution of primary healthcare physicians. Surgical treatment is possible in conditions of a specialised hospital. Further follow up of the infant demonstrated an increase in the diameter of the vessel appropriately for the growth of the body.

Conflict of interest: none declared.

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