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A rare case of coexistence of a patent ductus arteriosus and a congenital carotid arteriovenous fistula in a 7-year-old girl

Rzadki przypadek współistnienia przetrwałego przewodu tętniczego i wrodzonej przetoki tętniczo-żylnej szyjnej u 7-letniej dziewczynki

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Abstract

A 7-year-old girl, who underwent a hemodynamically significant patent ductus arteriosus closure at the age of 3 months, was diagnosed with a systolic gradient in the aortic lumen of approximately 40 mm Hg in Doppler examination, caused by prominence of the occluder. Additionally, an increase in the left ventricle and pulmonary artery sizes were established. Due to unclear symptoms, cardiac catheterization was performed, concluding that the implant was correctly positioned and did not obstruct blood flow, yet revealing features of increased left-right flow and pulmonary hypertension, suggesting the presence of an intracranial arteriovenous fistula. The performed computed tomography excluded a cerebral fistula and revealed an abnormal connection between the branch of the left external carotid artery and the left internal jugular vein. The fistula was interventionally closed with coils and no residual leakage was observed after the procedure. The girl was discharged home in a good condition.

Key words: arteriovenous fistula, carotid artery, patent ductus arteriosus, interventional treatment

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Introduction

Vascular fistulas, both congenital and acquired, are very rare (about 1% of all heart defects), appearing in the process of angiogenesis, abnormal arterio-arterial, arterio-venous or veno-venous connections, and may be located in various parts of the body. The most common fistulas are located in the systemic, coronary or intrapulmonary

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circulation. In all of the cases, the capillary circulation is bypassed [1, 2]. A systemic arterio-venous (A-V) fistula located in the brain (e.g., Galen's vein malformation) can lead to intracranial bleeding caused by a rupture of the thin wall of the fistula [3]. Large fistulas can lead to heart failure due to significant left-right shunt. Symptoms such as systolic-diastolic murmur above the fistula, rapid pulse, high systolic-diastolic pressure difference, hyperkinetic apical beat, gallop rhythm and tachycardia can be found. Doppler echocardiography, computed tomography and/or magnetic resonance imaging are essential in the diagnosis [4, 5]. The modern and safest method of treating fistulas is the interventional implantation of various endovascular devices into the lumen of the fistula [6]. In the case of very large intrapulmonary fistulas, cardiosurgical treatment is also considered [1, 6, 7].

Case report

A 7-year-old girl underwent a hemodynamically significant patent ductus arteriosus (PDA) closure with an Amplatzer Duct Occluder at the age of 3 months. Recent control echocardiography revealed left ventricular enlargement, dilatation of the pulmonary trunk and indentation of the implant into the aortic lumen, causing narrowing and flow velocity acceleration. The patient was admitted for diagnosis. Physical examination only exposed a soft systolic murmur at the left edge of the sternum. The echocardiographic image indicated a mild form of coarctation of the aorta, while the implant slightly extended into the aortic lumen. The Doppler examination at the isthmus site, revealed an acceleration of flow with a maximum gradient of 40 mm Hg, with no features of "coarctation-type" flow. Due to the reported chest pain that intensified during exercise, the patient was qualified for cardiac catheterization. During the procedure, direct measurements and angiography showed no evidence of aortic coarctation requiring dilatation, the implant was in the correct position and no leakage at the vascular level was observed (Figure 1), however, the examination revealed pulmonary hypertension (systolic blood pressure 39 mm Hg; mean 30 mm Hg). The hemodynamic image indicated the presence of an intracranial fistula however a contrast-enhanced computed tomography imaging of the head exposed no presence of a fistula in this region. Therefore, the area of the examination was extended to the neck and only then the A-V fistula was visualized in the subcranial region, between the branch of the left external carotid artery and the left internal jugular vein. A loud systolic-diastolic murmur was heard in the neck near the lower region of the left ear (Figure 2). The girl was qualified for fistula closure (Figure 3) and an interventional, effective closure was performed using coils (Figure 4).

Discussion

The simultaneous occurrence of two different congenital extracranial vascular fistulas in the same child, i.e., PDA and the carotid A-V fistula, is extremely rare, while iatrogenic fistulas acquired after catheterization of the internal jugular vein are more often diagnosed in the neck [7–10]. A fistula can be diagnosed clinically based on a characteristic systolic-diastolic murmur at the site of the fistula. Until

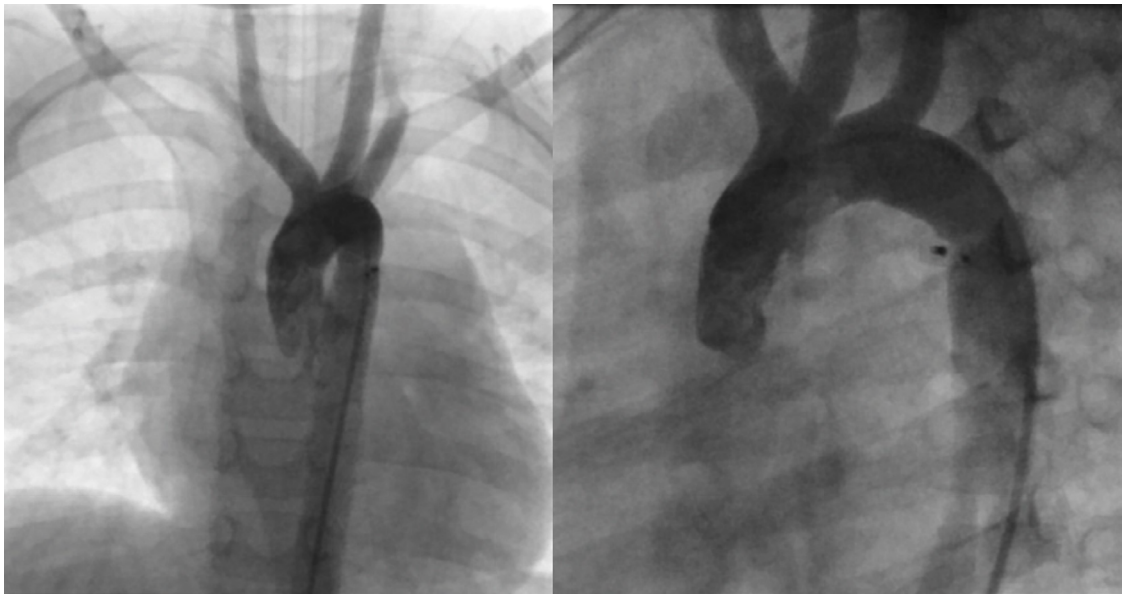

Figure 1. Aortography. Implant inside persistent ductus arteriosus in the correct position. Aortic coarctation was excluded

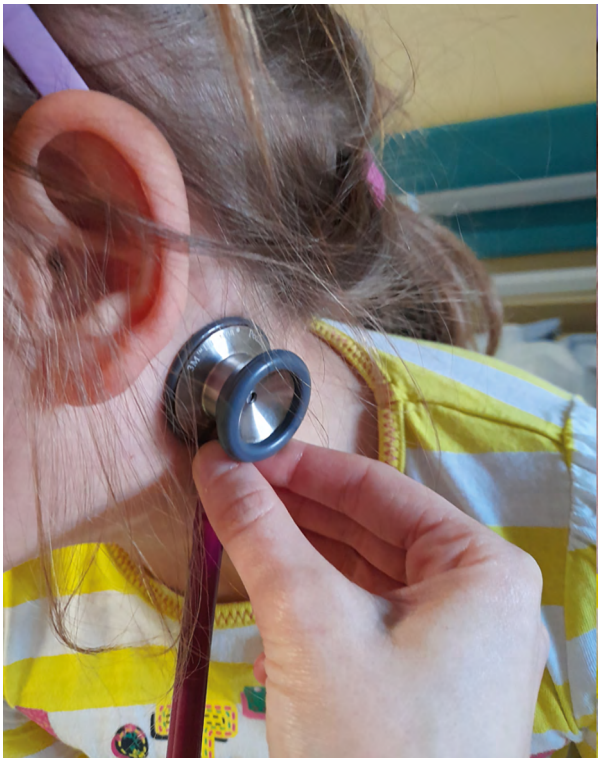

Figure 2. After placing the stethoscope in the area near the lower region of the left ear, a loud systolic-diastolic murmur was heard (photo with the consent of the patient and mother)

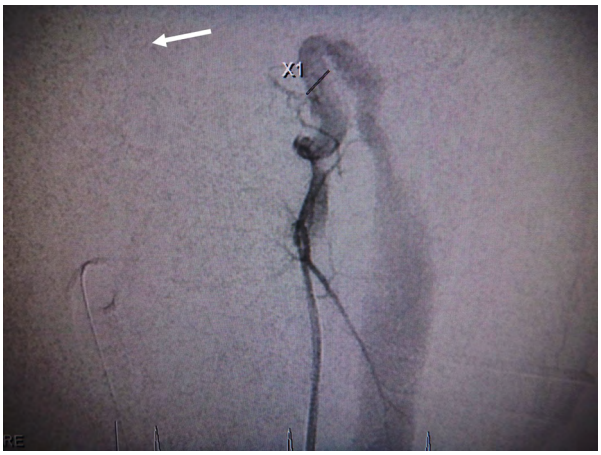

Figure 3. Arteriography. A catheter placed in the left external carotid artery. Visible arteriovenous fistula between the branch of the left external carotid artery and the left internal jugular vein (arrow)

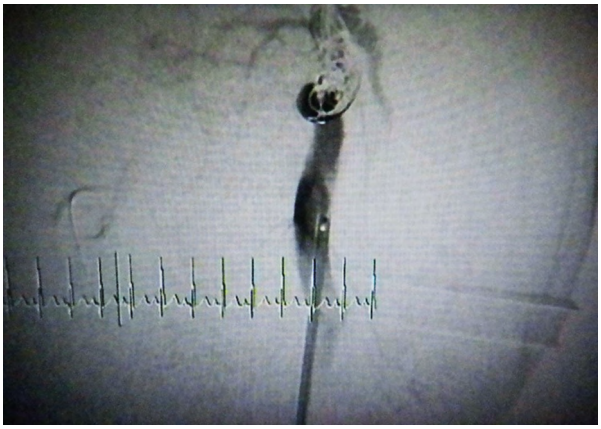

Figure 4. Arteriography. A catheter placed in the left external carotid artery. A fistula tightly closed with a coil occluder

the age of 7, the girl was in good condition and showed no disturbing symptoms until an abnormal image appeared in the aortic arch and PDA occluder implantation site examinations. It is possible to speculate, that if not for the cardiac catheterization performed to explain the causes of left ventricular enlargement, the vascular fistula in the neck would probably not have been diagnosed for a long time. On the other hand, the fistula could have been suspected after auscultatory examination, since one of the symptoms was a loud continuous systolic-diastolic murmur in the area of the lower pole of the left ear. An in-depth analysis of the course of treatment of the patient allowed to establish, that in some cases a more insightful auscultatory examination, such as auscultation of the cranio-jugular area, may expose symptoms of initially unexpected medical conditions.

Conclusions

Carotid arteriovenous fistulas in children are very rare and diagnosed at random. Hearing a continuous murmur of the fistula with a stethoscope may facilitate diagnosis. The treatment of choice is the interventional closure of the fistula.

Conflict of interest

None declared.

Funding

None.

Streszczenie

U 7-letniej dziewczynki, której w 3. miesiącu życia zamknięto implantem istotny hemodynamicznie przetrwały przewód tętniczy, w badaniu metodą Dopplera stwierdzono wpuklenie się okludera do światła aorty i jej przewężenie z gradientem skurczowym około 40 mm Hg. Ponadto powiększył się wymiar lewej komory oraz poszerzyła się tętnica płucna. Z powodu niejasnych objawów wykonano cewnikowanie serca — stwierdzono, że implant jest prawidłowo umieszczony i nie powoduje utrudnienia przepływu krwi. Wykluczono koarktację aorty, natomiast ujawniono cechy zwiększonego przepływu lewo-prawego z cechami nadciśnienia płucnego, sugerujące obecność wewnątrzczaszkowej przetoki tętniczo-żylnej. Po wykonaniu tomografii komputerowej wykluczono przetokę mózgową, uwidoczniono nieprawidłowe połączenie pomiędzy gałęzią lewej tętnicy szyjnej zewnętrznej a lewą żyłą szyjną wewnętrzną. Przetokę zamknięto interwencyjnie za pomocą cewek, po zabiegu nie zaobserwowano przecieku resztkowego. Pacjentkę wypisano ze szpitala w stanie dobrym.

Słowa kluczowe: przetoka tętniczo-żylna, tętnica szyjna, przetrwały przewód tętniczy, leczenie interwencyjne

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