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Cardiology in the Young (2015), **25**, 780–782
doi:10.1017/S1047951114001036

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Brief Report

Cerebral ischaemia after repair of coarctation of the aorta

Maria Gogou, Anastasia Keivanidou, Andreas Giannopoulos

2nd Pediatric Department, Aristotelian University of Thessaloniki, University General Hospital AHEPA, Thessaloniki, Greece

Abstract A 9-year-old boy, with a history of repair of severe coarctation of the aorta through balloon angioplasty 2 weeks ago, presented in the emergency paediatric department with symptoms consistent with transient cerebral ischaemia. MRI revealed an area of cerebral infarction in the right frontal lobe. Causes of cerebral ischaemia after aortic coarctation repair are briefly discussed.

Keywords: Coarctation of the aorta; stroke; cerebral ischaemia; children

Received: 13 February 2014; Accepted: 18 May 2014; First published online: 9 June 2014

Case report

A 9-year-old boy presented in the emergency paediatric department owing to symptoms of hypoesthesia of the left side of the head, and hypoesthesia and muscle weakness of the left upper limb. The episode took place 2 hours before his admission, lasted for 5 minutes, and since then the boy had been free of symptoms. The child had undergone a repair of severe coarctation of the aorta through balloon angioplasty 15 days ago and was administered metoprolol, captopril, and ranitidine. According to medical report, the angioplasty was performed under general anaesthesia, using a 5 Fr introducer sheath and a 0,035" floppy-tip guidewire. The guidewire was passed into the ascending aorta with its tip placed just above the coarcted segment. The coarctation was dilated by a balloon of size 12 mm/3 cm. An intravenous bolus of 100 IU/kg heparin was administered. The postoperative period was uncomplicated.

Physical examination and laboratory tests at the emergency department had no pathologic findings. The blood pressure was normal in all four extremities. A computed tomogram of the head performed at the time of admission was normal. An MRI of the brain was performed 3 days later, revealing a punctuate area of abnormal signal intensity, indicative of a recent ischaemic cerebral infarction. The lesion was located in the margin between the grey and white matter of the right frontal lobe, and there was no abnormal uptake from the cerebral structures (Fig 1). Moreover, a magnetic resonance angiography revealed a mild narrowing of the right internal carotid artery and its furcations, and also a mild narrowing of the initial segment of the right anterior cerebral artery, and of the A1 segment of the right middle cerebral artery (Fig 2).

The boy was also heterozygote for a mutation in the gene of factor V Leiden and had a lipoprotein a value of 270.5 mg/dl (reference range: ⩽30 mg/dl).

Cardiac dimensions and contractility were normal, whereas a mild aortic valve regurgitation was revealed in M-MODE two-dimensional echocardiography with colour Doppler. The mean residual pressure gradient in the region of angioplasty was 19 mmHg. Recommendations for a biannual cardiologic evaluation and regular control of lipidemic profile were given and the boy was discharged home with metoprolol and acetylsalicylic acid.

Coarctation of the aorta accounts for 5–7% of congenital heart diseases and often coexists with the bicuspid aortic valve and defects in the cerebral vessels, especially in the circle of Willis. Although it can be easily detected by femoral pulse palpation, it is often undiagnosed until late childhood, thus increasing the risk for cerebrovascular accidents, which are due to either the pathophysiology of the coarctation or the repair of this disorder.[1]

Correspondence to: M. Gogou, MD, Dimitriou Nika 44, Katerini 60 100, Greece. Tel: +0 030 697 890 5873; Fax: +0 030 235 107 4625; E-mail: mariaangogou@gmail.com

https://doi.org/10.1017/S1047951114001036 Published online by Cambridge University Press

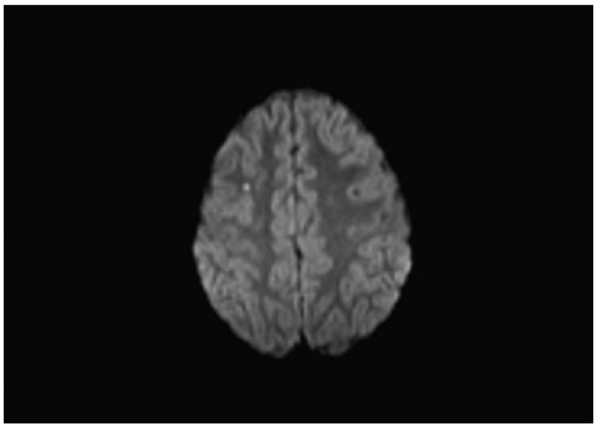

Figure 1.
The area of abnormal signal intensity, as it was revealed in the MRI of the brain.

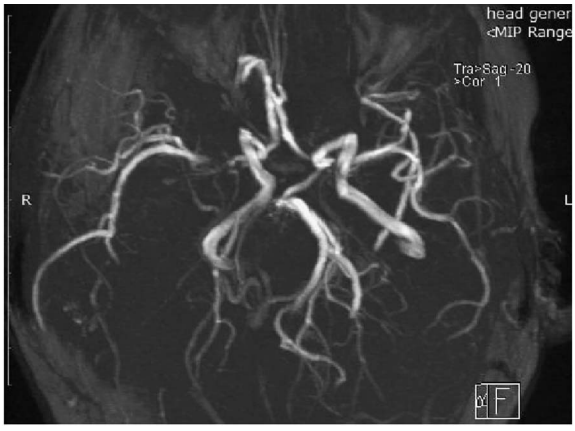

Figure 2.
A magnetic resonance angiography revealing a narrowing of the right internal carotid artery.

Subarachnoid or intracerebral haemorrhage can occur as a result of systemic hypertension and high blood pressure in the brain, especially in a background of defects of cerebral vessels. Young et al[2] have identified aortic coarctation as an uncommon cause of cerebral haemorrhage in four full- or near-term infants, even with moderate elevation of systolic blood pressure. Similarly, Opio et al[3] reported the case of a 14-year-old girl with coarctation of the aorta presenting as an acute haemorrhagic stroke, whereas Daghero et al[4] presented a 14-year-old boy with coarctation of the abdominal aorta, who had subarachnoid haemorrhage secondary to an aneurysm of the left posterior communicating artery.

With regard to repair of aortic coarctation as a cause of stroke, Brawn et al[5] presented the case of a 5-year-old boy who developed a left cerebellar infarction following repair of coarctation of the aorta by subclavian aortoplasty owing to ligation of a large left vertebral artery during surgery. Moreover, postoperative persisting hypertension has been proven to be relatively common even after successful repair of aortic coarctation and consists a major risk factor for the postoperative occurrence of cerebrovascular disease, especially haemorrhagic stroke. Its prevalence increases with age of repair, and most studies suggest that it is associated with decreased aortic compliance and abnormal neuroendocrine activation.[6] Benson et al[7] reported the case of a 10-month-old male child who developed post-procedural haemorrhagic cerebral infarction after balloon angioplasty for a recurrent aortic coarctation. Necropsy revealed a hypoplasia of the left posterior communicating artery of the circle of Willis.

Variations in cerebral blood flow are a probable mechanism of cerebral infarction in our patient; his cerebral parenchyma had been chronically exposed to high values of blood pressure because of coarctation of the aorta. The sudden reduction in cerebral blood flow after angioplasty could have led to disturbed blood supply in the cerebral structures, even resulting in ischaemic episodes. This effect is enhanced by the existence of defects in major cerebral vascular branches as well as by mutation of factor V Leiden and increased levels of lipoprotein a. Although elevated lipoprotein a and mutated factor V Leiden are independent risk factors for stroke in all ages, the close temporal relationship between repair of coarctation and ischaemic episode cannot be overlooked.

The possibility of embolism cannot be totally excluded, as post-catheterisation arterial thrombosis is not rare in children.[8] Ussia et al[9] have reported a case of paraplegia following balloon angioplasty of aortic coarctation in an infant, most likely due to embolism.

In general, cardiac disorders are associated with ~30% of strokes in childhood. Compared with children with stroke of other origin, children with ischaemic stroke due to congenital heart diseases are overall younger and have a higher prevalence of bilateral strokes and a greater tendency to haemorrhagic stroke.[10]

Broadly, children with aortic coarctation represent a high-risk population for stroke not only as a result of undiagnosed coarctation but also as a consequence of repair of this disorder. This fact underlies the importance of timely diagnosis of aortic coarctation and renders necessary the magnetic resonance angiography of cerebral vessels after diagnosis, as the presence of extensive cerebral vascular dysplasia may have adverse consequences on the outcome of surgical repair of this heart defect. Moreover, heightened clinical attention for stroke after repair of coarctation of the aorta, especially in children with late diagnosis, is required along with additional research for prevention of complications associated with repair of congenital heart diseases.

https://doi.org/10.1017/S1047951114001036 Published online by Cambridge University Press

Acknowledgement

None.

Financial Support

This research received no specific grant from any funding agency, commercial, or not-for-profit sectors.

Conflicts of Interest

None.

Ethical Standards

This report does not include any research involving human and/or animal experimentation.

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