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Case Report

Isolation of left subclavian artery with reversal of neurological and hemodynamic abnormalities after percutaneous closure

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ABSTRACT

Isolation of left subclavian artery is a rare congenital anomaly. In this abnormality, the left subclavian artery arises from the homo-lateral pulmonary artery rather than from aorta. This condition is often diagnosed by angiography and treated by surgery. The authors present a case, which had vertebro-basilar insufficiency, subclavian steal phenomenon and pulmonary plethora. All these clinical signs disappeared by a simple percutaneous intervention.

1. Introduction

Isolation of left subclavian artery is invariably associated with right aortic arch. The incidence of right aortic arch is 0.05% in the general population.[1] Isolation of left subclavian artery is the rarest, accounting for 0.8% of all right aortic arch[2] abnormalities. In 60% of such cases, isolation of left subclavian artery is associated with complex congenital heart disease especially Tetralogy of Fallot.[3] As an isolated defect it may be clinically silent. Its presence is suspected when there is lung flooding in a cyanotic baby or there is pulmonary plethora and a weak left radial pulse or even critical limb ischemia.

2. Case report

A 4-year-old girl was brought to our hospital because of recurrent episodes of respiratory tract infection. She demonstrated features of both physical (weight – 8.5 kg and height – 92 cm) and mental under development. She was unable to stand and had nasal regurgitation of food. She also had nasal intonation of voice.

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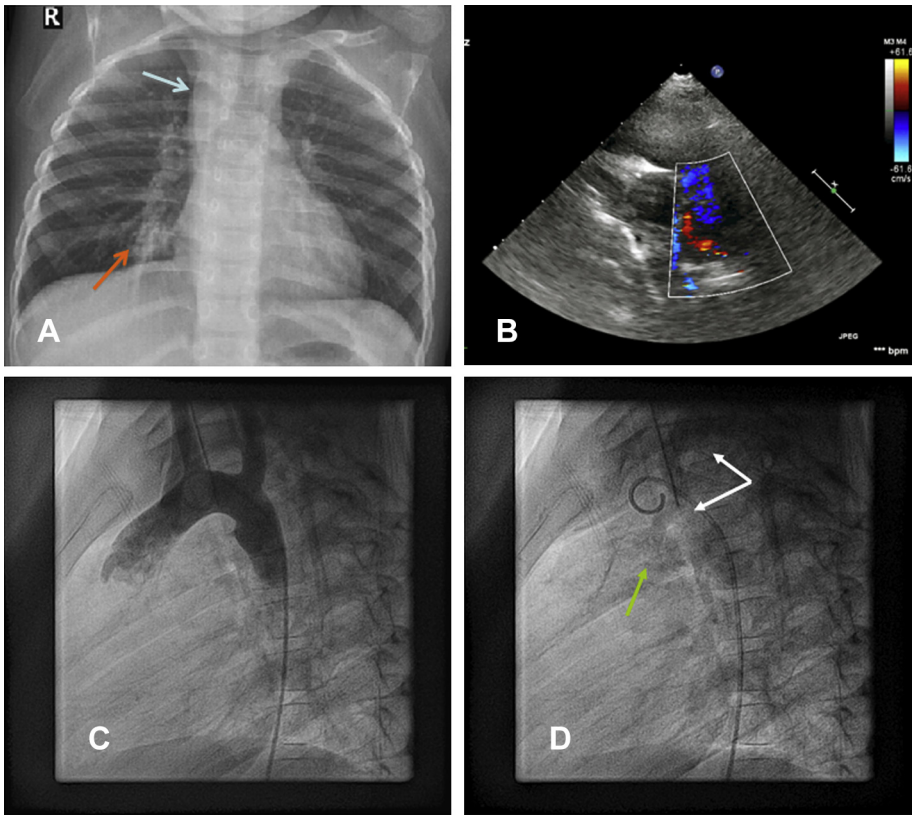

Fig. 1 – A – Chest X-ray in postero-anterior view shows pulmonary plethora (orange arrow) and right aortic arch (blue arrow) but no cardiomegaly with cardiothoracic ratio of 0.5. B – Transthoracic echocardiogram in parasternal short axis view shows continuous flow in pulmonary artery seen at the origin of left pulmonary artery. C – Aortic arch angiogram in left lateral view shows there is no evidence of patent ductus arteriosus. D – Aortic arch angiogram in dead lateral view shows filling of pulmonary artery (green arrow) from left vertebral and subclavian artery (white arrow).

Left upper limb systolic pressure was 25 mmHg lower than that of the right arm.

Auscultation revealed a continuous murmur over the second left intercostals space near the sternum. Her chest X-ray showed pulmonary plethora and a right aortic arch (Fig. 1A). Echocardiogram in parasternal short axis view demonstrated continuous flow in pulmonary artery (Fig. 1B), but there was no coarctation of aorta. The clinical suspicion was isolation of left subclavian artery or a patent ductus with left subclavian ostial stenosis.

Although the arch angiogram in the left lateral view did not show any duct patency, late filling of pulmonary artery through an abnormal vascular structure from the neck was seen; this is suggestive of systemic-to-pulmonary artery shunting (Fig. 1C and Movie 1). Aortic root angiogram confirmed the diagnosis of right aortic arch and isolation of left subclavian artery. The first branch was the left common carotid, the second was the right common carotid and the last one was the right subclavian artery with absent left subclavian artery (Fig. 2A). A dilated and tortuous right vertebral artery was seen, which was supplying the left vertebral and subclavian arteries though the circle of Willis and in turn communicating with the left pulmonary artery (Fig. 2A, B). Therefore, it was inferred that the patient had dual steal phenomena; one was at the level of vertebro-basilar system causing vertebro-basilar insufficiency and the other was at the level of systemic-to-pulmonary artery, contributing to significant left-to-right shunt, explaining recurrent lower respiratory tract infection and lower systolic blood pressure in left arm.

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The shunt was calculated to be 2.5:1, and the duct was sized to be 3.6 mm at the tightest point. Hence, it was decided to close the shunt. Both surgical as well as percutaneous intervention options were discussed with the parents. Because of multiple co-morbidities, parents were reluctant for surgery. Hence an 8–6 Cera flex patent ductus arteriosus occluder (Lifetech Scientific limited) device was deployed at the narrowest point from pulmonary arterial side. A complete exclusion of the left subclavian from the pulmonary artery and improved flow through the left subclavian artery were achieved instantaneously as shown in (Fig. 2C, D and Movie 2).

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During follow up, there was no further episode of respiratory tract infection or regurgitation of food. She has gained