

Atypical aortic pseudocoarctation in a pediatric patient

Pseudocoartación aórtica atípica en un paciente pediátrico

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ABSTRACT

Aortic pseudocoarctation, a rare congenital aortic anomaly, mimics coarctation but typically lacks significant hemodynamic impact. We present a six-year-old male evaluated for systemic arterial hypertension. Diagnosis involved echocardiography revealing preductal coarctation and a wide patent ductus arteriosus, confirmed by cardiac CT angiography showing atypical double narrowing and angulation. Surgical coarctectomy and patent ductus arteriosus ligation via left thoracotomy were successfully performed. The critical role of advanced imaging in differentiating this condition is crucial.

Keywords: aortic arch, congenital heart, cardiac surgery.

Aortic pseudocoarctation, an infrequent congenital aortic anomaly, involves the elongation, stenosis, and kinking of the aorta, primarily at the isthmus. Despite its shared clinical presentation with aortic coarctation, pseudocoarctation typically does not produce a substantial gradient in the descending aorta or significant hemodynamic sequelae. Given this overlap and crucial distinction, comprehensive imaging techniques –foremost among them cardiac CT angiography and catheterization– are paramount for accurate differential diagnosis.¹ The specific case we report was an incidental discovery during the management of systemic arterial hypertension.

RESUMEN

La pseudocoartación aórtica, una anomalía aórtica congénita poco frecuente, simula una coartación, pero generalmente carece de impacto hemodinámico significativo. Presentamos el caso de un niño de seis años evaluado por hipertensión arterial sistémica. El diagnóstico se realizó mediante ecocardiografía, que reveló coartación preductal y un conducto arterioso persistente ancho, confirmado mediante angiografía cardíaca por TC, que mostró un doble estrechamiento y angulación atípicos. Se realizó con éxito una coarctectomía quirúrgica y una ligadura del conducto arterioso persistente mediante toracotomía izquierda. La imagenología avanzada es crucial para diferenciar esta afección.

Palabras clave: arco aórtico, cardiopatía congénita, cirugía cardíaca.

CASE DESCRIPTION

This is the case of a six-year-old male presenting with an eight-month history of systemic arterial hypertension, referred to our service for evaluation and to rule out cardiac pathology.

On physical examination, we noted rhythmic heart sounds with a grade II/VI ejection murmur at the accessory aortic focus, along with a grade II/VI systolic murmur in the posterior hemithorax and physiological splitting of the second heart sound. There was no hepatomegaly, but pulses in the lower limbs were diminished.

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A transthoracic echocardiogram revealed a preductal aortic coarctation with a maximum gradient of 25 mmHg and a wide patent ductus arteriosus, measuring 9 mm from the pulmonary mount. Subsequent cardiac CT angiography of the heart and great vessels confirmed a double area of narrowing, exhibiting both counterclockwise and posterior clockwise angulation (*Fig. 1*), in addition to a wide patent ductus arteriosus. This imaging effectively ruled out other aortic arch pathologies.

The patient was presented at a medical-surgical session and accepted for coarctectomy and ligation of the arterial duct.

Via a left lateral thoracotomy, dissection was performed in layers to access the thoracic cavity, exposing the aortic arch, ductus arteriosus, left common carotid artery, left subclavian artery, brachiocephalic trunk, and descending thoracic aorta. Both the juxtaductal aortic coarctation and the ductus arteriosus were clearly visualized. We then performed proximal and distal clamping of the ductus arteriosus, sectioning both ends and ligating them with 5-0 polypropylene. Vascular clamps were applied to the aortic arch and distal aorta, followed by an extended end-to-end anastomosis towards the arch using 4-0 polypropylene, achieving good filling with no evidence of leaks. During the procedure, we observed an atypical angulation of the aortic arch, consistent with the CT angiography findings (*Fig. 2*). The aortic clamping time was 17 minutes, and the total operative time was 180 minutes. Postoperative echocardiography showed no significant residual gradient.

The patient was extubated in the operating room and discharged 72 hours after the operation. He is currently under routine follow-up, remains complication-free, and maintains blood pressure within normal limits.

COMMENT

Pseudocoarctation of the aorta constitutes an exceedingly rare congenital anomaly, precipitated by the embryonic compression of the middle third of the seventh dorsal aortic segment. This segment, being anomalously shorter than anticipated, culminates in an elongated aortic arch that convolutes around the ductus arteriosus.^{2,3} Consequently, this elongation engenders a distinct gap between the origin of the left carotid artery and that of the left subclavian artery.⁴

It can be concomitantly associated with an array of other cardiac anomalies, including aortic coarctation, bicuspid aortic valve, ventricular and atrial septal defects, as well as patent

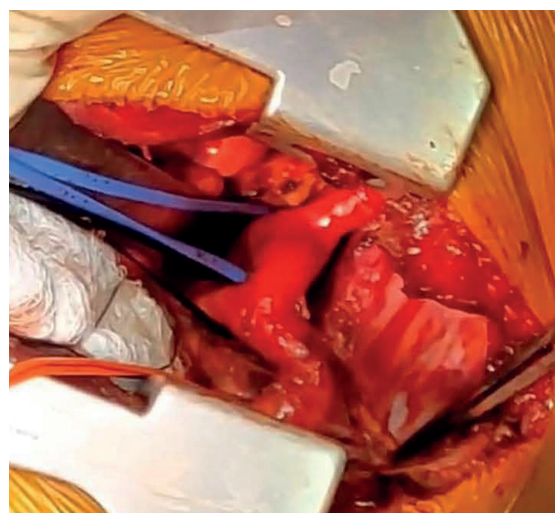
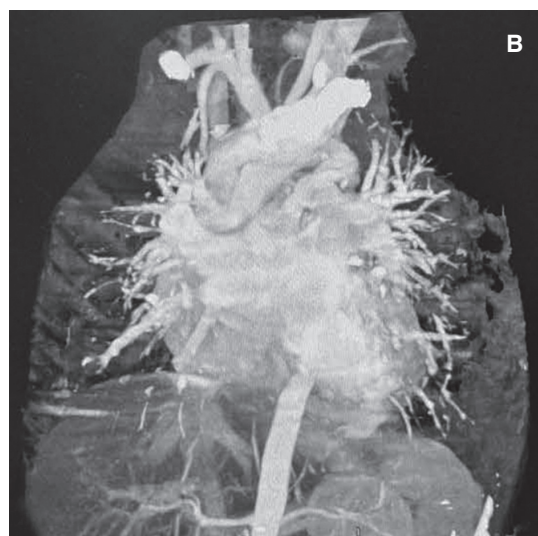
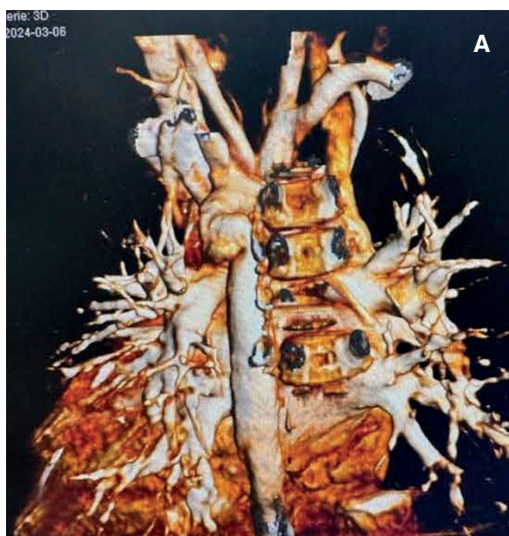


Figure 2: Intraoperative image showing the area of atypical angulation.

Figure 1:

Angiotomography showing a double zone of narrowing with clockwise and counterclockwise angulation.



ductus arteriosus,⁵ the latter of which constitutes a pertinent association in our particular case. While collateral flow is commonly encountered in instances of aortic coarctation, pseudocoarctation is distinguished by the notable absence of such collateral flow.⁶ Furthermore, pseudocoarctation may manifest as an integral component of more complex syndromes, such as Turner syndrome, Noonan syndrome, and Hurler syndrome.^{2,7}

Clinically, pseudocoarctation manifests predominantly as resistant hypertension,⁸ a presentation exemplified by the patient in our case, whose initial clinical manifestation was systemic arterial hypertension. The management paradigm for asymptomatic patients with pseudocoarctation of the aorta is generally considered to be conservative in nature. However, in the presence of overt clinical symptoms or significant concomitant cardiac abnormalities, surgical intervention ought to be duly considered.^{2,4}

CONCLUSIONS

In culmination, it is imperative to underscore that pseudocoarctation of the aorta should not be regarded as a benign pathological entity. This exceedingly rare condition exhibits a pronounced clinical similitude to genuine coarctation, thereby mandating fastidious diagnostic acumen. The exemplar case we proffer herein was ascertained

incidentally during the therapeutic management of systemic arterial hypertension.

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