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Case Report

# Renovascular hypertension secondary to Takayasu's arteritis in a child: Case report

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ABSTRACT

*Introduction:* Takayasu arteritis (TA) is a chronic inflammatory arteriopathy of unknown etiology that affects the aorta and its branches. Inflammation leads to arterial stenosis, thrombosis, and aneurysm formation. Management strategies for TA include medical therapy and revascularization procedures after inflammation is controlled.

*Presentation of the case:* We report the case of a 12-year-old female patient, who presented with dyspnea and growth retardation. Initial clinical and radiological evaluations revealed hypertension and congestive heart failure. After controlling the inflammatory phase of the disease and blood pressure partially (with three antihypertensive agents), the patient underwent percutaneous balloon angioplasty of both renal arteries, with angiographic and clinical success. At the 2-year follow-up, she presented with re-stenosis of the right renal artery and progression of the supra-renal aortic stenosis. She was subjected to a second balloon angioplasty of the right renal artery and aortic balloon angioplasty.

*Discussion:* After controlling the inflammatory phase of the disease, stenotic and/or aneurysmal lesions can be addressed. Percutaneous revascularization of renal arteries is reasonable for patients with hemodynamically significant renal artery stenosis.

*Conclusion:* TA with renal involvement must be considered as an etiologic factor for secondary hypertension in young patients, even if there is no blood pressure difference between the upper extremities. In this case, renal and aortic stenosis were performed with low perioperative morbidity.

## 1. Introduction

Takayasu's arteritis (TA) is a chronic inflammatory arteriopathy of unknown etiology that affects the aorta and its major branches, and sometimes the pulmonary arteries [1–3]. The disease is prevalent in Asians and women under 40 years [2,3].

Although the etiology is unknown, TA is characterized by segmental and patchy granulomatous inflammation of the aorta and its major branches. Inflammation leads to arterial stenosis, thrombosis, and aneurysm formation. Irregular fibrosis of the blood vessels can occur due to chronic vasculitis, which sometimes leads to massive intimal fibrosis (fibrosis of the inner sections of the blood vessels). The signs and

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symptoms depend on the affected vessels [2].

Management strategies for TA include medical therapy with corticosteroids and/or immunosuppressive agents during the active (inflammatory) phase. Revascularization procedures are usually undertaken after the inflammation is controlled (chronic stage). Revascularization of the affected organ can be achieved by open surgery, balloon angioplasty, or stenting [2,4].

Isolated involvement of the abdominal aorta and its branches (Lupi-Herrera type II class) is relatively rare; it has been reported in only about 12% of patients with TA [5]. This case follows the 2020 SCARE guidelines for reporting surgical cases [6].

## 2. Presentation of case

A 12-year-old female patient presented with dyspnea and growth retardation. Initial clinical and radiologic evaluations identified hypertension and congestive heart failure, characterized by sustained high blood pressure associated with dyspnea and an increase in the cardiac silhouette on chest radiography.

The patient underwent computed tomography angiography and magnetic resonance angiography; these showed narrowing of the abdominal suprarenal aorta and both renal arteries. Laboratory analysis revealed markedly elevated erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) levels, and creatinine/blood urea nitrogen levels; all other rheumatologic work-up examinations were negative (anti-neutrophil cytoplasmic antibody, antinuclear factor, anti-DNA, extractable nuclear antibody, anti-Ro antibody, anti-cardiolipin antibody, and circulating immune complexes). Therefore, the most reasonable etiology for arterial disease was TA (Lupi-Herrera type II).

After controlling the inflammatory phase with corticosteroids (prednisone 1 mg/kg/day) and an immunosuppressive agent (methotrexate 15 mg/m2/week) and controlling the blood pressure partially with three antihypertensive agents, the patient underwent arteriography, which showed subocclusive lesions on both proximal renal arteries and a 50% stenotic lesion of the suprarenal abdominal aorta (Fig. 1). The patient underwent balloon angioplasty of the renal arteries, with immediate angiographic success (Fig. 2); the procedure was accomplished with general anesthesia. Through right femoral artery access, both renal arteries were treated with a 5 × 20 mm semi-compliant angioplasty balloon. After this treatment, the patient's blood pressure was adequately controlled with an antihypertensive drug.

During follow-up 2 years after the initial percutaneous treatment, the patient presented with renal scintigraphy, showing reduced size and impaired glomerular function of the right kidney. She required treatment with a second antihypertensive drug, despite supposedly adequate control of the underlying inflammatory disease (normal ESR and CRP). A new angiographic examination showed right renal artery re-stenosis and progression of the previous aortic suprarenal stenosis (Fig. 3).

A second percutaneous transluminal balloon angioplasty was performed to re-treat the right renal artery (5 × 20 mm semi-compliant balloon) and to treat the aortic stenosis (12 × 60 mm semi-compliant balloon), with immediate technical/angiographic success (Fig. 4). This second procedure was also accomplished under general anesthesia by right femoral artery percutaneous access. The patient was discharged on the first postoperative day with double antiplatelet aggregation and was followed-up in the outpatient clinic of Vascular Surgery and Rheumatology without further complications, until the third postoperative month.

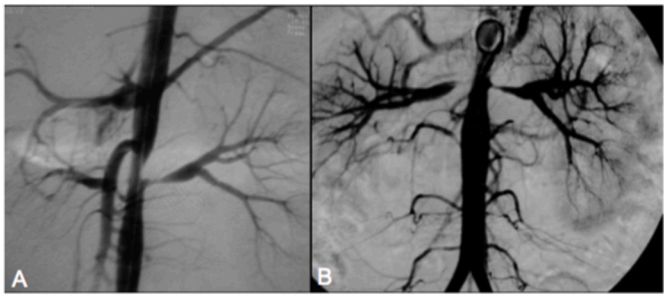


**Fig. 1.** Abdominal aortic angiogram. Subocclusive lesions of both renal arteries and focal suprarenal aortic stenosis. Left anterior oblique view (A) and posteroanterior view (B).

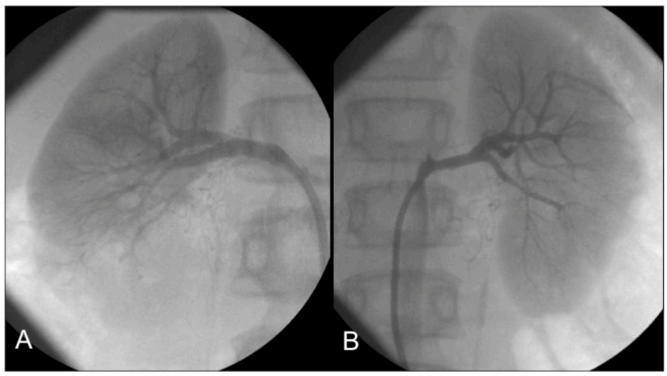


**Fig. 2.** Selective renal angiogram. Post balloon angioplasty immediate angiographic result of the right renal artery (A) and left renal artery (B).

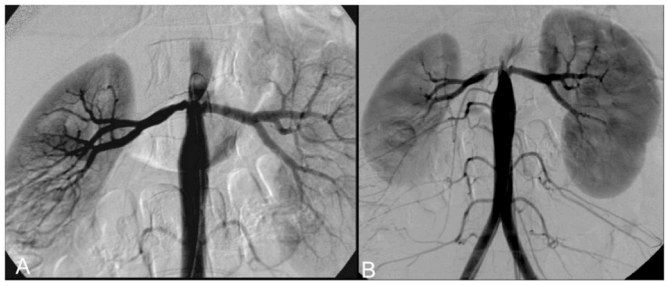


**Fig. 3.** Abdominal aortic angiogram. Restenosis of the right renal artery (A) and progression of the suprarenal aortic stenosis (B).

## 3. Discussion

TA is a chronic granulomatous inflammatory arteriopathy that primarily affects the large elastic arteries such as the aorta and its major branches. Most patients develop symptoms between the ages of 20 and 40 years (with no trend by region), which makes the present case uncommon as the patient was 12 years old [7]. The diagnosis was based on angiographic findings and a complete laboratory work-up so that other vasculitis, vascular infections, fibromuscular dysplasia, and inflammatory idiopathic syndromes could be ruled out.

According to the classification proposed by Ueno and modified by Lupi-Herrera, the involvement of the abdominal aorta and both renal arteries is characteristic of Lupi-Herrera type II [5,8]. A retrospective study by Lupi-Herrera et al. reported an incidence of 12% for Lupi-Herrera type II (total of 107 patients studied) [5]. Renal artery involvement occurs in 20–50% of patients and is the most common bilateral ostial lesion, leading to secondary hypertension, usually with difficult medical control, as in this case [9].

Notably, renovascular hypertension (RVHT) occurs because renal artery stenosis causes hypoperfusion of the juxtaglomerular apparatus, resulting in exaggerated secretion of renin and high blood aldosterone levels, which eventually leads to water and salt retention and sustained high blood pressure. In studies from Korea, Thailand, and China, TA was the most common cause of RVHT in children and young adults [10]. It

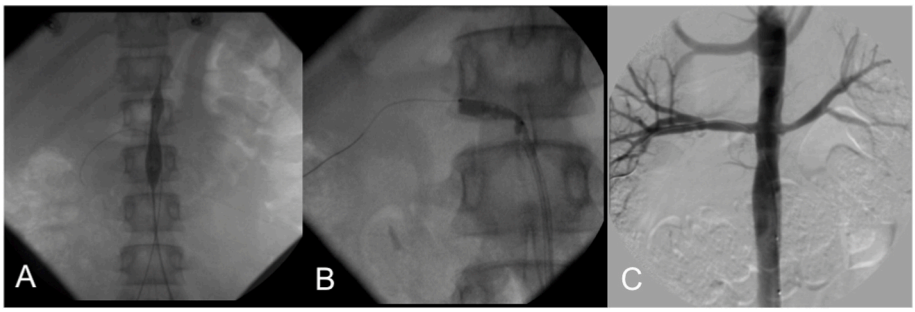


**Fig. 4.** Balloon angioplasty of the aortic lesion (A). Balloon angioplasty of the right renal lesion (B). Immediate post treatment nonselective aortic angiogram (C).

poses a double threat: severe hypertension (and its complications) and progressive renal insufficiency, as shown in this case. At only 12 years of age, she presented with signs of heart failure and arterial disease progression, even after optimal medical treatment.

After controlling the inflammatory phase of the disease, stenotic and/or aneurysmal lesions can be addressed; roughly, treatment indications for the arterial residual lesions are similar to those of lesions with other etiologies. Percutaneous revascularization of the renal arteries is reasonable for patients with hemodynamically significant renal artery stenosis and accelerated hypertension, resistant hypertension, and malignant hypertension with or without decreased renal function [11].

To our knowledge, the first percutaneous transluminal angioplasty in TA with renal involvement was performed by Saddekni et al. [12]. Since then, it has been the main treatment modality for renal artery proximal lesions, with good results in pediatric patients with TA [13]. In a Korean study, patency rates after renal artery angioplasty in TA patients were 90% after 1 year and 50% after 10 years, demonstrating the need for close follow-up of these patients for re-intervention for de novo lesions [14]. Stable disease activity and treatment with corticosteroids and immunosuppressive agents have been found to aid in the maintenance of arterial patency [7].

## 4. Conclusion

TA with renal involvement must be considered as an etiologic factor for secondary hypertension in young patients, even if there is no blood pressure difference between the upper extremities (which is the most frequent clinical presentation of the disease). Endovascular treatment can be performed with low periprocedural morbidity to achieve primary patency and assisted primary patency, as shown in the present case.

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## Ethical approval

As the manuscript is not a research study, we only have the patient consent for writing and others forms of publication. Also, the ethical approval for this case reports has been exempted by our institution.

## Consent

Written informed consent was obtained from the patient's family for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## Author contribution

Ricardo Magalhães, Juliana Matos, Juan Rodriguez and Priscilla Campelo contributions to conception, design, collected the patient details and wrote the paper. Paulo Diniz, José Souza and Leonardo Cavalcante made contributions to patient management. Leonardo Cavalcante, Priscilla Campelo and Marcos Velludo critically revised the article. All authors read and approved the final manuscript.

## Registration of research studies

The manuscript is a case report, not considered a formal research involving participants.

## Guarantor

Ricardo Filipe Souza Magalhães.

## Declaration of competing interest

We do not have any conflicts of interests.

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