Percutaneous transluminal angioplasty in pediatric patients with Takayasu arteritis: comparison of initial and long- term results of interventions on aorta and non-aortic vessels

Takayasu arteritli pediatrik hastalarda perkütan balon anjiyoplasti: Aort ve non-aortik damarlarda erken ve uzun dönem girişim sonuçların karşılaştırılması

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Takayasu arteritis (TA) is a chronic inflammatory condition of the aorta and other major vessels that usually results in stenosis, occlusion, dilatation, or the formation of aneurysm. The studies on long-term results of percutaneous transluminal angioplasty (PTA) in pediatric patients with TA are limited (1-3). Furthermore, there is no study that compared PTA results of large and medium vessels. Herein we report 15 pediatric TA cases treated with PTA for stenotic lesions of the aorta, renal arteries, celiac truncus and superior mesenteric artery. The aim of this study is to compare initial and long- term results of PTA in large- and medium-sized vessel.

Between August 1992 and January 2007, 15 pediatric TA cases with significant stenosis (≥60% stenosis of vessel diameter) of the aorta and/or non-aortic vessel involvement were treated with PTA. Fifteen cases (9 girls, 6 boys; age range 2.5 months- 18 years; median age, 12 years) were included.

Informed consent was obtained from all parents. Although unilateral access was preferred, in two cases, bilateral femoral artery access was used with kissing balloon technique to reduce trauma to the common femoral artery (Table 1). For PTA procedures of the aorta, while inflating the balloon in aorta, renal arteries were protected by keeping the guide-wire in them (3). In cases with severe stenosis (case 6 and case 12), "graded dilatation" was performed to reduce the risk of rupture (Table 1). It means dilatation by gradually increasing balloon diameter.

If residual stenosis was less than 40% during the immediate post-PTA arteriogram, the procedure was considered technically successful. Clinical results were based on symptoms, pulses and blood pressure values and were judged as cured, improved or failed. The procedure was considered clinically successful if the clinical results were rated as cured or improved (3).

During follow up period, symptomatic patients with suspicious imaging findings were evaluated with diagnostic angiography whether the presence of recurrence (stenosis rate ≥50%).

The paired sample t-test, independent samples t-test and the Pearson's Chi-square test (Epi Info 2000 CDC, Atlanta, USA) were used for deciding about significance of differences for mean pre- and post-PTA stenosis, technical success of the procedures, rate and time of recurrences, primary patency rate (continued patency without subsequent intervention) and secondary patency rate (continued patency after any necessary intervention) between two groups.

During the initial PTA procedures, 13 of the 15 stenotic aorta segments, and 23 of the 26 stenotic non-aortic vessel segments were successfully dilated (Fig.1, 2). In case 4, poor clinical result was observed due to technically unsuccessful dilatation of both the aorta and renal arteries. Case 1 died as the result of an arrhythmia due to hypertrophic cardiomyopathy within 24 hours of PTA. All the remaining cases were clinically successful (Table 1).

The follow-up period was 4-168 months (mean: 53.7±65.88 months). Repeated procedure was performed in 6 recurrent cases (Table 1).

Overall, 12 of the 15 stenotic segments of the aorta and all the non-aortic vessels were successfully dilated. Due to the small number of cases, statistical evaluation was not performed for clinical results: however, the outcomes are presented in Table 1. The comparison of PTA results between groups are listed in Table 2.

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Table 1. The characteristics of patients, and details of the procedures with initial and follow-up results

Patient				Stenosis		Procedure		Initial result		Follow-up			
Case no	Age	Sex	Symptom and Diagnosis	Location	Length of the segment, cm	Entry route	Balloon size, mm	Technical success (for each segment)	Clinical success*	Follow up time, month	Recurrence (involved segment with time of recurrence)	Adequate dilation (for each segment)	Clinical outcome*
1	2.5 m	M	HT Hypertrophic Cardiomyopathy	SAA IAA	4.2 3.8	Ax. A	6x40 6x40	+ +	Exitus	-	-	-	Exitus
2	11 m	M	HT, Dyspnea, Hypertrophic Cardiomyopathy CHF	PDTA- DDTA** SAA	10.6 2.5	Ax. A	5x100 5x100 6x20	+ + +	Improved	33	PDTA -DDTA SAA 9th and 33rd month	- -	Failed (operated)
3	3 y	M	HT	R RA L RA	0.8 1.2	CFA	4x20 5x20	+ +	Cured	6	-	+ +	Cured
4	6 y	F	HT LLC	SAA IAA R RA L RA	5 3 1.6 1.8	CFA	4,5,6x40 4,5,6x40 4x20 3.5x20	- - - -	Failed	168	SAA IAA R RA L RA 1st, 9th in all segments, 48th and 72nd in the aorta	+ + + +	Cured
5	8 y	F	HT	R RA L RA	1.2 1.5	CFA	4x20 4x20	+ +	Cured	6	-	+ +	Cured
6	10 y	F	HT Dyspnea, LLC	DDTA –SAA**	6	CFA	6x60 8x60	+ +	Improved	154	-	+ +	Improved
7	10 y	F	HT	R RA L RA	0.9 1.3	CFA	4x20 4x20	+ +	Cured	36	-	+ +	Cured
8	12 y	M	HT LLC	SAA-IAA** R RA	5 1.6	CFA	6x60 5x20	+ +	Improved	36	-	+ +	Improved
9	13 y	M	HT	L RA R RA L RA	1.3 0.8 1.2	CFA	4x20 4x20 4x20	+ + +	Improved	8	L RA 5th month	+ +	Improved
10	14 y	F	HT	R RA L RA	2 1.7	CFA	4x20 4x20	- +	Improved	4	R RA 3rd month	+	Improved
11	15 y	F	HT	DDTA SAA R RA R RA*** L RA	3.8 3 2 1.4 1.9	CFA	12x40 12x40 4x20 4x20 5x20	+ + + + +	Cured	36	-	+ + + + +	Cured
12	15 y	F	HT ULC Abdominal angina	SAA L RA TC SMA	3.6 1 1.2 1.2	CFA	12x40 4,5,6x20 6x20 6x20	+ + + +	Improved	156	-	+ + + +	Improved
13	16 y	F	HT ULC	R RA L RA	0.8 1.3	CFA	5x20 5x20	+ +	Cured	12	R RA L RA 6th month	+ +	Cured
14	16 y	M	HT	R RA L RA	1.4 1.6	CFA	4x20 4x20	+ +	Cured	6	-	+ +	Cured
15	18 y	F	HT ULC LLC	IAA R RA L RA	3 2 1.7	CFA	12x40 4x20 5x20	+ + +	Improved	144	R RA L RA 12th month	+ + +	Improved

Ax. A-axillary artery, CFA- common femoral artery, CHF-congestive heart failure, CT- celiac truncus, DDTA- distal descending thoracic aorta, HT- hypertension, IAA- infrarenal abdominal aorta, LLC- lower limb claudication, LRA- left renal artery, m-months, PDTA- proximal descending thoracic aorta, RRA- right renal artery, SAA- suprarenal abdominal aorta, SMA- superior mesenteric artery, y - years, ULC- upper limb claudication
*Cured: resolution of the symptoms, ability to palpate previously impalpable arterial pulses, normal blood pressure values without the use of antihypertensive drugs. Improved: improvement of the symptoms, ability to palpate previously impalpable arterial pulses, even if not sufficiently strong, normal blood pressure values with antihypertensive drug use or at least a 15% reduction in diastolic blood pressure without the use of antihypertensive drugs. Failed: no change or worsening of symptoms, arterial pulses, and blood pressure values

: continuous stenosis, *: Accessory R RA

Table 2. Procedural characteristics

Variables	Aortic group	Non-aortic group	p*
Initial procedures			
mean pre-PTA stenosis, %	74.7	84.0	0.020
mean post-PTA stenosis, %	28.7	22.7	0.336
Initial technical success rate, %	86.7	87.5	0.866
Repeated procedures			
mean pre-PTA stenosis, %	64.3	76.7	0.022
mean post-PTA stenosis, %	31.4	24.8	0.145
Overall technical success rate, %	80	100	0.086
Follow-up			
mean rate of recurrence	1.08	0.35	0.043
mean time to recurrence	14 months	1.6 months	0.008
Primary patency rate, %	53.3	43.3	0.533
Secondary patency rate, %	66.7	100	0.004

Data are presented as ratios (percentages) and means
* Independent samples t test and Pearson's Chi square test
PTA - percutaneous transluminal angioplasty

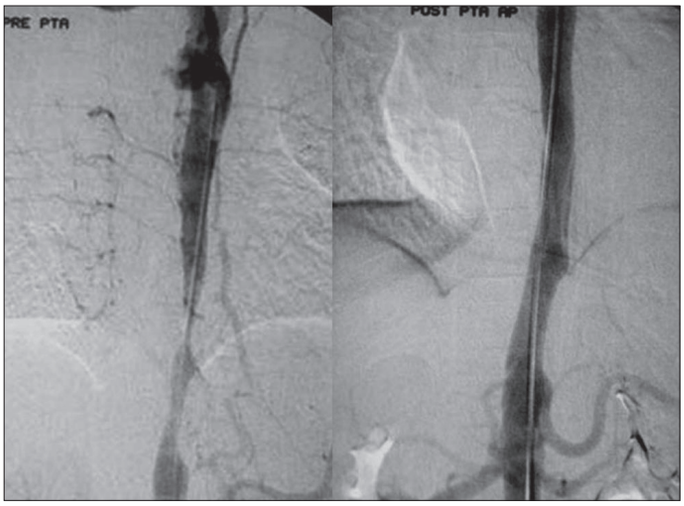

Figure 1. Antero-posterior view of pre- and post-PTA aortogram. The collateral vessels significantly diminished after adequate dilation (case no 6)
PTA - percutaneous transluminal angioplasty

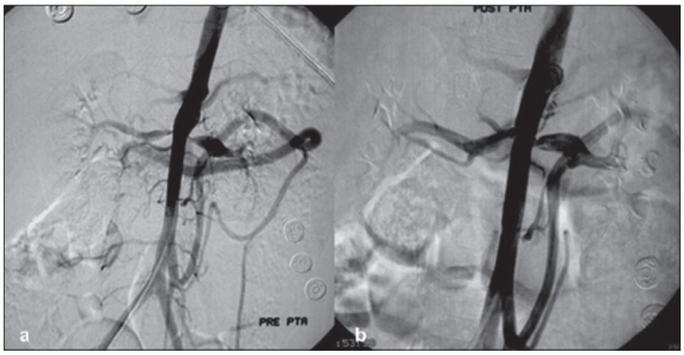

Figure 2. The significant stenosis of bilateral renal artery and adequate dilation with minimal residual stenosis is seen. Also note the post-stenotic dilation on the left side (case no 3)

Small intimal flaps were seen in case 1 and in case 2. Because the flaps were too small and were not flow restricting, stent implantation was not performed. The death of youngest case within 24 hours was the only major complication.

Up to now, there have been few renal angioplasty series limited to pediatric cases reported (1, 2, 4-6). The largest series was reported by Casalini et al. (4), followed by Tyagi et al. (1). Successful dilation was reported in 94.4% and in 88.6% of cases, respectively. We achieved success in 83.3% of cases during the initial PTA. Not many studies have been published on dilatation of aorta in pediatric patients. The two large series reported by Tyagi et al. (3) and Saxena et al. (7) achieved success in 92.7% and in 88.2% of cases at the initial PTA, respectively. We achieved success in 75% of cases. It was reported that patients with stenoses of the aorta extending across the renal arteries posed special problems. When dilating aorta, occlusion of the renal arteries is possible if dissection extends into the origins of the renal arteries (3). We are in agreement with Tyagi et al. (1) that keeping a wire in the renal artery while inflating the balloon across them is a good therapeutic approach to avoid occlusion of the renal arteries.

Despite the high initial success rate, the disease recurs in a substantial number of the patients treated. It was reported that more than 20% residual stenosis appears to increase the risk of restenosis of renal arteries (8) and long eccentric lesions do not respond to dilatation as well as short concentric lesions in aorta (9, 10). When comparing the two groups; we found that recurrence occurred earlier in non-aortic vessels, but more often in aorta. We do not know the reason of the difference. The alteration in amount of constituents of the vessel media layer may play a role.

It is difficult to draw definitive conclusion due to a small number of patients included as a main limitation. We deem our results preliminary and it should be reviewed with larger samples.

According to our results, PTA is an excellent treatment, with high technical success for both aortic and renal artery stenoses of TA. Although technical success is similar, the long-term course is different in two groups; recurrence occurs more often in the aorta, whereas earlier in the renal arteries. Re-dilated renal arteries remain more often patent than re-dilated aortic segments. Because of repeatability of the procedure, it should be considered as the initial treatment in pediatric patients with TA.

Conflict of interest: None declared.

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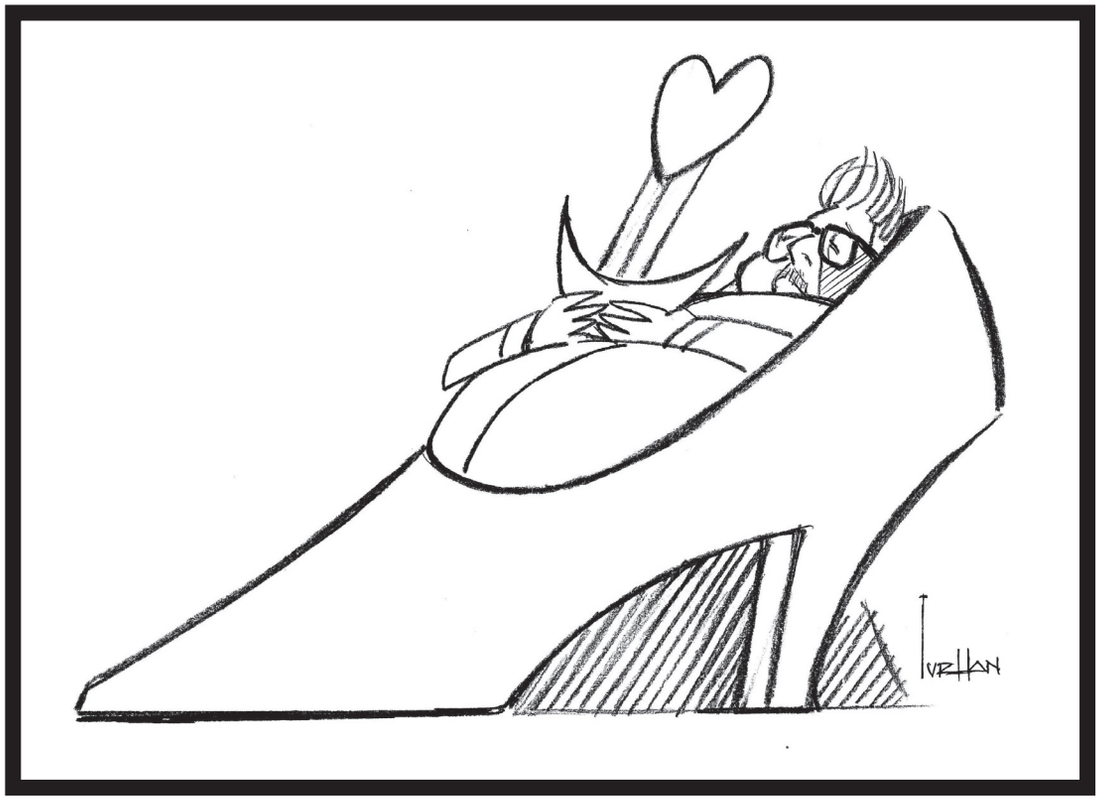

Turhan Selçuk ustaya saygı ile

Sayın Ruhan Selçuk'un izni ile