EVALUATION OF ARTERIAL ANATOMY IN CONGENITAL CLUBFOOT WITH COLOR DOPPLER ULTRASOUND

JOSÉ ANTONIO PINTO[1], FRANCESCO CAMARA BLUMETTI[2], LUIZ ALBERTO NAKAO IHA[3], MARCOS KIYOSHI TERASAKA[4], HENRIQUE SODRÉ[5], AKIRA ISHIDA[6]

SUMMARY

Objective: This investigation intended to evaluate anterior and posterior tibial arteries at the ankle joint level in congenital clubfoot, by using color Doppler ultrasound (CDU). Material and Method: Twenty patients with idiopathic clubfoot were selected, from which 18 had unilateral involvement and two had bilateral involvement. Of the 18 patients with unilateral clubfoot, 16 went through surgical treatment and the other two were submitted to conservative treatment with serial casting. Of the bilateral cases, one patient was treated surgically and the other was treated with serial casting. All patients were clinically and radiographically assessed. We used the functional rating as described by Lehman. Then, CDU was applied bilaterally at the ankle joint level, trying to identify both posterior and anterior tibial arteries. Results: In our present series of 20 cases with idiopathic clubfoot, in just one patient we could not identify the anterior tibial artery at the ankle joint level. In 12 patients who have had their arterial flow speeds and diameters measured by UDC, a positive correlation was found between functional level and anterior tibial artery diameter. No statistically significant differences were found between both flow speed and diameter of anterior tibial artery of the normal side, when compared to the affected side (in patients with unilateral disease). Conclusion: In our sample, we could not find any significant differences in arterial morphology and flow speed between the normal and the affected side. Furthermore, we noticed that the better the clinical result of clubfoot correction, the larger the diameter of anterior tibial artery in affected feet.

Keywords: *Clubfoot; Ultrasonography doppler color; Congenital abnormalities; Arteries.*

Citation: *Pinto JA, Blumetti FC, Iha LAN, Terasaka MK, Sodré H, Ishida A. Evaluation of arterial anatomy in congenital clubfoot with color doppler ultrasound. Acta Ortop Bras. [on-line]. 2008; 16(5):270-4. Available at URL: http://www.scielo.br/aob.*

INTRODUCTION

Congenital clubfoot (CCF) is a generic term used to describe a complex congenital condition of the foot, characterized by strong equinism, cavism, varusing, and adduction. There are four major kinds of CCF: postural, neurogenic, syndromic and idiopathic. In the present study, we will address the idiopathic kind only.

Its average incidence ratio is 1:1000 live births, being less common among Asians and more frequently seen in Hawaiians, Polynesians and Maoris. It is twice more common in males, and bilateral in 50% of the cases. When it is unilateral, the left foot is most commonly affected[1].

This is the most common congenital defect of feet, although its pathogenesis is still unclear.

Concerning etiology, Hippocrates defined CCF as a change originated from a vicious intrauterine positioning. Many authors have suggested other theories for its genesis, including the theory of vascular hypo development, but none of those can be proven. In the last decade, Kawashima and Uhthoff showed the existence of a shutdown on foot development, preventing its spontaneous correction, which would theoretically perpetuate deformities[2].

Anatomically, we observe a number of bone, muscular, and vascular abnormalities[1,3,4,5]. Several authors conducted arteriographic studies and found an association between CCF and hypoplasia or early anterior tibial artery end.[6-9]. In our environment, Sodré and cols, using the same methodology, found a deficiency of the anterior tibial artery in 90% of the cases[9].

Some previous studies, using the assessment with continuous-wave doppler in CCF patients, could not detect differences on the prevalence of vascular changes when compared to normal populations[10,11]. A study by Schwartz et al[12] suggested the possibility of using color doppler ultrasound (CDU) to assess artery anatomy in CCF. More recently, Katz et al[13]. used the CDU to assess the arterial anatomy of the CCF, concluding that the deficiency of the foot dorsal artery (defined as the presence of retrograde flow) was more common among patients with CCF than in control subjects.

This controversy has encouraged us to conduct a comparative study of the arterial flow on ankles by using the CDU in CCF patients. For a better analysis of outcomes, we especially assessed patients with unilateral condition and age group above 4 years old, with definite gait. We also correlated vascular changes found with feet functional degree.

MATERIALS AND METHODS

This study was approved by UNIFESP-EPM's committee of ethics in research, under the number 1343/06. Twenty patients have been selected with diagnosis of idiopathic CCF, being 18 males (90%), and two females (10%). Two patients (10%) were bilaterally affected, while 18 (90%) had the unilateral condition, totaling

Study conducted by the Discipline of Pediatric Orthopaedics and Traumatology – "Paulista" Medical School, Federal University of São Paulo.
Correspondences to: Rua: Borges Lagoa, 783 – 5o Andar – Vila Clementino – São Paulo – Brasil – CEP: 04038-032

1. Associate Professor and Clinical Head of the Discipline of Pediatric Orthopaedics, Department of Orthopaedics and Traumatology, UNIFESP.
2. Orthopaedic Doctor, Discipline of Pediatric Orthopaedics, Department of Orthopaedics and Traumatology, UNIFESP
3. Orthopaedic Doctor, Discipline of Pediatric Orthopaedics, Department of Orthopaedics and Traumatology, UNIFESP
4. Radiologist, Department of Imaging Diagnosis, UNIFESP
5. Associate Professor, Discipline of Pediatric Orthopaedics, Department of Orthopaedics and Traumatology, UNIFESP
6. Chairman and Head of the Discipline of Pediatric Orthopaedics, Department of Orthopaedics and Traumatology, UNIFESP

Received in 06/21/07 approved in 09/17/07

22 feet. Of the 18 patients with unilateral CCF, 16 (80%) were submitted to surgical treatment, and two (20%) to conservative treatment with serial plastered casts. Age group ranged from 5 to 12 years old, with an average of 7.45 years. Of the 18 patients with unilateral condition, the left foot was involved in 13 cases (72.2%), and the right foot was involved in five cases (27.8%). Sixteen patients (80%) were regarded as non-Caucasians and four were regarded as Caucasians (20%).

Previously to baseline evaluation, an informed consent term was handed to patients' caregivers on which the whole procedure to be performed was described. Anamnesis data (name, gender, age, ethnicity, affected side, plastered cast use, previous surgeries, presence of pain, functional restraint, and orthosis use) as well as information about physical examinations (ankle dorsiflexion, calcaneus position, forefoot appearance, flexor tendons function, and subtalar movement) were collected and transferred to a standardized patient file. X-ray images employed in this part of the assessment were the same as previously obtained during routine medical follow-up.

Lehman's criteria were employed[14] to record any relevant data concerning clinical and X-ray aspects, so that we could have objective values for our assessment (Chart 1). These criteria are classically described for CCF postoperative evaluation. We classified functional outcomes according to the values recommended by the author, with outcomes scoring 85-100 being regarded as excellent, 70-84 good, 60-69 fair, and <60 as poor (Figure 1).

Chart 1 - *Lehman's criteria for congenital clubfoot assessment*

1) Ankle dorsiflexion (passive)	
( ) > 90	15
( ) 90	5
( ) < 90	0
2) Subtalar movement (passive)	
( ) > 10	0
( ) <10	5
( ) Stiff	0
3) Calcaneus position at orthostatism	
( ) 0-5 valgus	10
( ) > 5 valgus	5
( ) varus 0	
4) Forefoot appearance	
( ) Neutral 10	
( ) < 50 abd/adduction	5
( ) > 50 abd/adduction	0
5) Gait	
( ) Normal 10	
( ) Doesn't walk on calcaneus	-2
( ) Doesn't walk on tips	-2
( ) Flatfoot gait	-4
6) Radiographic measures	
T-C Index	
(sum of TC angle at AP and P)	
( ) 40	5
( ) < 40	0
T-1 MTT Angle	
( ) 10	5
( ) 15	10
7) Shoes	
( ) Normal – no complaint	5
( ) Normal – with complaint	2
( ) Inner sole or orthosis	0
8) Function	
( ) No restraint	15
( ) Occasional restraint	8
( ) Frequent restraint	0
9) Pain	
( ) Never	10
( ) Occasionally	5
( ) Frequently	0
10) Flexor tendons	
( ) Total function	5
( ) Partial function	3
( ) No function	0

Figure 1 - *A 9 year-old patient with CCF, 8 years postoperatively, with Lehman index of 85.*

The color doppler ultrasound test was performed on the same day of clinical evaluation. During the test, we attempted to identify anterior and posterior tibial arteries at the level of ankle joint, always bilaterally. In 12 of these patients, values for flow and anterior and posterior tibial artery were obtained for later comparison.

The device employed in the test was the Envisor model (Philips) with 5 - 10 MHz linear transducer with correction angle of 60º for velocimetric analysis and cross sectional measurements of arteries gauge at the level of ankle joint (Figure 2). All tests were performed by the same radiologist. As we didn't find in literature any normality parameters for this age group, we used the unaffected contralateral limb as control.

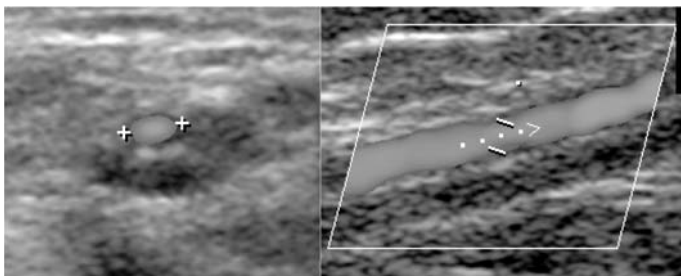

Figure 2 - *Illustration of the diameter and arterial flow measurement technique using CDU*

In order to compare the correlation between anterior and posterior tibial arteries'gauge and flow, we created two coefficients. The first coefficient was named as FluxAP, which consists of the ratio between values obtained from anterior tibial artery flow over posterior tibial artery flow values. The other coefficient was named as CalAP, which represents the ratio between anterior and posterior tibial arteries'gauge values. These coefficients were employed only in cases of unilateral conditions where the contralateral side could serve as control for comparison purposes.

Data obtained from both study phases were compiled and submitted to statistical analysis for establishing the conclusions. Data were correlated by Pearson's method and by the Student's t-test for paired samples.

RESULTS

The key clinical data of the patients assessed in this study are summarized on Table 1.

All patients in this series were initially treated with serial plastered casts using Kite's methodology, which is routinely employed by the original service, up to a mean age of 12 months. Sixteen patients were submitted to surgical treatment upon conservative therapy failure. Of these, three were submitted to two surgical interventions, while other 2 were submitted to three interventions each. Only 4 patients had not been submitted to surgical treatment.

Concerning functional level of the 22 feet assessed according to Lehman's criteria, the average value obtained was 60.4 (± 20.35), ranging from 16 to 90. The distribution of these values can be found on Table 2.

All patients were submitted to analysis of anterior and posterior tibial arteries, bilaterally, with the U/S. In 12 of these patients, flow and gauge were measured by arterial doppler-velocimetry. As in this case the condition was bilateral, we totaled 13 feet submitted to objective measurement tests (Table 3).

The mean anterior tibial artery gauge in the present study was 1.7 ± 0.15mm on the normal side, and 1.44 ± 0.13mm on the affected side. The mean values for anterior tibial artery flow obtained on the normal side were 56.61 ± 6.15cm/s, and, on the affected side, 47.72 ± 7.44mm.

Values for FluxAP and CalAP coefficients were obtained on the 11 patients with unilateral CCF submitted to flow and gauge measurements. These values are shown on Table 4.

Table 1 – *Distribution of results concerning number of patients, age, gender, bilateral or unilateral disease, affected side, time of plastered cast use and postoperative time of the first surgery.*

Nr	Age	Gender	Bilateral/ Unilateral	Side	Cast (months)	PO time (years)
1	10	M	Unilateral	Right	12	8.5
2	7	M	Unilateral	Left	24	5
3	8	M	Unilateral	Right	12	3
4	5	M	Bilateral	Both	12	-
5	11	M	Unilateral	Left	12	9.5
6	6	F	Unilateral	Left	8	5
7	9	M	Unilateral	Left	11	8
8	7	M	Unilateral	Left	12	6
9	7	M	Unilateral	Right	9	-
10	7	M	Unilateral	Left	14	1
11	5	M	Unilateral	Left	5	4
12	6	M	Unilateral	Right	8	5
13	5	M	Unilateral	Left	6	4
14	8	M	Unilateral	Right	12	7
15	7	F	Unilateral	Left	12	6
16	8	M	Bilateral	Both	24	5
17	5	M	Unilateral	Left	9	-
18	7	M	Unilateral	Left	6	6
19	9	M	Unilateral	Left	19	8
20	12	M	Unilateral	Left	10	-

Table 2 – *Distribution of values obtained according to Lehman's criteria*

Lehman	Frequency	%
< 60	9	40.91%
60-69	8	36.36%
70-84	3	13.64%
85-100	2	9.09%

Table 4 - *Distribution of FuxAP and CalAP coefficients' values on the normal and affected sides.*

Nr	Normal Side		Affected Side	
	FluxAP	CalAP	FluxAP	CalAP
1	1.103	1.182	0.946	1.222
2	0.985	0.950	1.067	0.739
3	0.986	1.167	0.961	0.640
4	0.822	0.800	0.921	0.700
5	0.812	0.750	0.867	0.762
6	1.512	0.783	1.335	0.955
7	0.991	0.762	0.368	0.696
8	0.719	1.000	0.467	0.857
9	0.469	0.722	0.433	0.667
10	0.818	1.176	1.233	1.067
11	0.468	0.524	0.428	0.474

Table 3 – *Distribution of the results concerning patient number, Lehman's criteria, measurements of flow (cm/s) and gauge (mm) of anterior and posterior tibial artery on affected and normal sides. *Patients with bilateral involvement. †Patient in whom posterior tibial arteries have been visualized but could not be measured for flow. ND: not detected. P: Present.*

Nº	Lehman	Anterior Tibial art. Affected side		Posterior Tibial art. Affected side		Anterior Tibial art. Normal side		Posterior Tibial art. Normal side	
		Flow	*Gauge*	*Flow*	*Gauge*	*Flow*	*Gauge*	*Flow*	*Gauge*
1	90	53.8	2.2	56.9	1.8	63.1	2.6	57.2	2.2
2	70	92.5	1.7	86.7	2.3	87.7	1.9	89	2
3	43	64.8	1.6	67.4	2.5	69.9	2.1	70.9	1.8
4	63	30.4	0.7	33	1	28.1	0.8	34.2	1
5	80	62.4	1.6	72	2.1	60.3	1.5	74.3	2
6	85	96.1	2.1	72	2.2	75.6	1.8	50	2.3
7	85	33.7	1.6	91.5	2.3	73.1	1.6	73.8	2.1
8	63	60	1.8	128.5	2.1	61.5	2.1	85.5	2.1
9	64	19.7	1.2	45.5	1.8	30.3	1.3	64.6	1.8
10	51	24.9	1.6	20.2	1.5	37.7	2	46.1	1.7
11	53	21.7	0.9	50.7	1.9	35.5	1.1	75.9	2.1
12*	21D	14.2	0.8	P†	P†	-	-	-	-
	16E	46.2	1	P†	P†	-	-	-	-
13	90	P	P	P	P	P	P	P	P
14	53	ND	ND	P	P	P	P	P	P
15	66	P	P	P	P	P	P	P	P
16*	65D	P	P	P	P	-	-	-	-
	70E	P	P	P	P	-	-	-	-
17	29	P	P	P	P	P	P	P	P
18	53	P	P	P	P	P	P	P	P
19	63	P	P	P	P	P	P	P	P
20	56	P	P	P	P	P	P	P	P

DISCUSSION

Concerning gender, we could not establish any correlation with arterial anatomy, considering that only two female patients have been assessed. Concerning age, a positive significant correlation ($p < 0.01$) was found for anterior tibial artery flow (Pearson = 0.31) and gauge (Pearson = 0.27) on the affected side. However, when we assess the normal side, no correlation was found for age, flow (Pearson = 0.04) and gauge (Pearson = 0.008). The natural development of the anterior tibial artery in children happens in the same pace with the normal limb growth. As oppositely to expected, the above results suggest, in our sample, a stronger arterial development of the affected feet than in normal feet. It is possible that, in the treated CCF cases, a positive response occurs to orthostatism and to an improved foot positioning, enabling a favorable evolution of the arterial pattern. However, a prospective study would be required with serial CDU during the development of CCF patients in order to confirm these findings.

We also saw a positive correlation, although at a lower degree, between postoperative time, flow (Pearson = 0.24) and gauge (Pearson = 0.14), both statistically significant ($p < 0.01$). It is possible that when a plantigrade foot is formed and gait age starts, the muscular development of the end also stimulates vascular growth. Although there is a direct correlation between patient's age and postoperative time, a doubt remains as for how arterial flow and gauge would increase if no deformity correction occurred. The correlation between arterial flow and gauge on operated patients and those with uncorrected congenital clubfoot deserves further studies.

We noticed in this sample a large percentage of cases with fair and poor outcomes according to Lehman's criteria. Most of these patients were initially treated at their original services, and often submitted to late surgical treatment. When we assess the correlation between the value achieved with Lehman's criteria and the anterior tibial artery flow on the affected side, we can see a trend towards a positive correlation (Pearson = 0.41), which was not statistically significant ($p = 0.06$). On the other hand, when we assess gauge over Lehman's values, such correlation (Pearson = 0.67) was statistically significant ($p < 0.01$).

By comparing the range of flow and gauge values for anterior tibial artery between the normal and the affected side through Student's t-test for paired samples, we didn't see any statistically significant difference ($p = 0.11$ for flow and $p = 0.02$ for gauge).

Concerning FluxAP and CalAP coefficient values, only in one case, these values were lower than 50% (patient nr. 11) (Table 4). However, the same pattern was found on patient's normal side. We didn't find statistically significant differences in the Student's t-test between FluxAP values on normal and affected side ($p = 0.23$). Similarly, no statistical significance was found when we compared the CalAP values on both sides, although they have somehow shown a trend ($p = 0.05$).

These data are contrary to previous arteriographic studies reported by literature [6-9]. However, most of those studies were conducted in children below the age of two, before being submitted to surgical treatment. The study by *Greider and cols* involved children aged 1.3 to 13.5 years, but all of these were serious and untreated cases[8]. The study by Katz et al with arterial CDU in CCF has also found anterior tibial artery deficiencies in up to 45% of the cases[13]. Nevertheless, that study was conducted on 6-12 month-old children, before gait age.

With improved foot positioning after treatment, an almost normal circulatory pattern might be developed with a smaller number of biasing factors. This could somehow explain the positive correlation between a better clinical outcome and a wider anterior tibial artery diameter. The correlation of deformity degree and anterior tibial artery deficiency has already been pointed out in literature by Edelson and Husseini[10], in a study using continuous-wave doppler. In this author's sample, not so many cases of anterior tibial pulse were found as it is reported by literature.

Several previous studies advocated a vascular theory for CCF genesis. Hootnick et al.[15] had already proposed some factors against this theory, such as: a portion of the patients with CCF had a normal arteriographic pattern of the lower limbs; many other malformations are associated to the same flow pattern; 3.7 to 12% of the normal population may present deficiencies or absence of anterior tibial artery[16]; standalone reports in literature report absent posterior tibial artery[17-20]. Notwithstanding, the final conclusion of the study was that an etiologic significance existed for the vascular hypothesis of CFF genesis.

The vascular changes reported by literature may not play an established causal role. The difference on circulatory pattern would then be only another else within the spectrum of abnormalities observed in CCF. Therefore, it is likely that an anterior tibial artery deficiency in CCF represents only a temporary functional change, which would be resolved with the development of an appropriately treated limb.

In order to improve the representativeness and accuracy of the study, a larger patient sample would be required. In addition, supplementary studies with more specific imaging tests, such as angioresonance, could provide important contributions to the knowledge about vascular changes and their role in CCF genesis.

CONCLUSION

On the sample assessed here, no significant change was found for arterial morphology and flow on patients with CCF. In addition, we noticed that the better the clinical outcome of CCF correction, the larger the gauge of the anterior tibial artery on affected feet.

ACKNOWLEDGEMENTS

We acknowledge Dr. Fernando Soma for his support and guidance on radiological aspects of this study.

REFERENCES

1. Morrissy RT, Weinstein S. Ortopedia pediátrica de Lovell e Winter. 5ª ed. Barueri: Manole; 2005.
2. Kawashima T, Uhthoff HK. Development of the foot in prenatal life in relation to idiopathic club foot. J Pediatr Orthop. 1990;10:232-7.
3. Hootnick DR, Levinsohn EM, Randall PA, Packard DS Jr. Vascular dysgenesis associated with skeletal dysplasia of the lower limb. J Bone Joint Surg Am. 1980; 62:1123-9.
4. Hootnick DR, Levinsohn EM, Crider RJ, Packard DS Jr. Congenital arterial malformations associated with clubfoot. Clin Orthop Relat Res. 1982; (167):160-3.
5. Hootnick DR, Packard DS Jr, Levinsohn EM, , Lebowitz MR, Lubicky JP. The anatomy of a congenitally short limb with clubfoot and ectrodactyly. Teratology. 1984; 29:155-64.
6. Polo GV, Ruiz GP. Reporte preliminary al hallazgode la ausencia vascular en enfermus con pies equino cavo varo aducto congênito. Rev Ortop Latinoam. 1968; 8:27-34.
7. Ben-Menachem Y, Butler JE. Arteriography of the foot in congenital deformities. J Bone Joint Surg Am. 1974; 56:1625-30.
8. Greider MD, Stiff SJ, Gerson P, Donovan NM. Arteriography in clubfoot. J Bone Joint Surg Am. 1982; 64:837-40.
9. Sodre H, Bruschini S, Mestriner LA, Miranda F Jr, Levinsohn EM, Packard DS Jr et al.. Arterial abnormalities in talipes equinovarus as assessed by angiography and the Doppler technique. J Pediatr Orthop. 1990;10:101-4.
10. Edelson JG, Husseini N. The pulseless clubfoot. J Bone Joint Surg Br. 1984; 66:700-2.
11. Stanitski CL, Ward WT, Grossman W. Noninvasive vascular studies in clubfoot. J Pediatr Orthop. 1992;12:514-7.
12. Schwartz RA, Kerns D, Fillinger M. Color doppler imaging for assessment of arterial anatomy in congenital skeletal foot deformity. In: Simons GW. editor. The clubfoot: the present and a view of the future. New York: Springer-Verlag; 1994. p.59-62.
13. Katz DA, Albanese EL, Levinsohn EM, Hootnick DR, Packard DS Jr, Grant WD et al. Pulsed color-flow Doppler analysis of arterial deficiency in idiopathic clubfoot. J Pediatr Orthop. 2003; 23:84-7.
14. Lehman WB, Atar D, Grant AD, Strongwater AM. Functional rating system for evaluation of long-term results of clubfoot surgery. In: Simons GW. editor. The clubfoot: the present and a view of the future. New York: Springer-Verlag; 1994. p.114-116.
15. Hootnick DR, Packard DS Jr, Levinsohn EM, Wladis A. A vascular hypothesis for the etiology of clubfoot. In: Simons GW. editor. The clubfoot: the present and a view of the future. New York: Springer-Verlag; 1994. p.48-59.
16. Sarrafian SK. Anatomy of the foot and ankle. Philadelphia: Lippincott; 1983.
17. Kitziger K, Wilkins K. Absent posterior tibial artery in an infant with talipes equinovarus. J Pediatr Orthop. 1991; 11:777-8.
18. Quillin SP, Hicks ME. Absent posterior tibial artery associated with clubfoot deformity: an unusual variant. J Vasc Interv Radiol. 1994; 5:497-9.
19. Schwering L, Ruppert R, Reichelt A. Aplasia of the posterior tibial artery in a child with idiopathic clubfoot. Orthopade. 2003; 32:437-8.
20. Dobbs MB, Gordon JE, Schoenecker PL. Absent posterior tibial artery associated with idiopathic clubfoot. A report of two cases. J Bone Joint Surg Am. 2004; 86:599-602.