

REVIEW

## Popliteal Artery Entrapment Syndrome in Children: Experience With Four Cases of Acute Ischaemia and Review of the Literature

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### WHAT THIS STUDY ADDS

Popliteal artery entrapment syndrome (PAES) is a well known entity in adults and has been widely documented in the literature. This anatomical abnormality is uncommon in paediatric practice and is often an unheralded diagnosis, the disease often being discovered at the stage of complications. In this report clinical and surgical experience of PAES in children with acute ischaemia is detailed. In addition, data were analysed from the literature with emphasis on the clinical severity on admission. The various diagnostic strategies and the outcome of treatment in paediatric patients were assessed. This paper gives interesting information about an extensive review of PAES cases in children not reported before.

**Objective:** Popliteal artery entrapment syndrome (PAES) is an uncommon anatomical anomaly, frequently described in adults. The most common symptom is claudication. Acute limb ischaemia (ALI) in children is rare, but it may evolve and lead to limb loss or lifelong complications. Clinical and surgical experience of PAES in children is reported. Data from the literature are analysed in order to assess the severity of this disease and to identify the factors characterising the diagnosis and the outcome of treatment in paediatric patients.

**Methods:** Four children (aged 7–16 years) were referred with ALI due to PAES. Among the 439 articles reporting cases of PAES, 55 patients under 18 years of age were the focus. The PAES cases were classified according to the Love and Whelan classification modified by Rich.

**Results:** Data from 79 children (106 limbs, 27 bilateral PAES) were collected and analysed. Type I PAES was present in 41 (39%), Type II in 23 (22%), Type III in 24 (23%), Type IV in 12 (11%), and Type V in two (2%) limbs. A functional PAES was present in one patient bilaterally. In two cases, the type of PAES was not reported. Claudication occurred in 68 cases (64%), and ALI in 19 (18%). In 60 cases (57%), revascularisation with or without myotomy was required; myotomy alone was performed in 41 cases (39%).

**Conclusions:** Symptomatic PAES in children should be considered a severe condition requiring urgent investigation in order to avoid any delays in the treatment. Early diagnosis and treatment are essential to prevent serious complications. The long-term outcomes of surgical treatment with the correction of the anatomical anomaly and vascular reconstruction are satisfactory with a low complication rate.

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### INTRODUCTION

Popliteal artery entrapment was first described by Stuart in 1879<sup>1</sup> as an abnormal anatomical relationship between the popliteal artery and local musculo-tendinous structures caused by an anomaly of embryological development. The term popliteal artery entrapment syndrome (PAES) was introduced by Love and Whelan in 1965.<sup>2</sup> PAES is caused by

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compression of the popliteal artery by the surrounding musculo-tendinous structures.

The most common symptom of PAES is lower leg pain with calf claudication. This mostly occurs in young adults (30–40 years) with well developed muscles, such as soldiers and athletes.<sup>3,4</sup> However a number of reports have described cases of children presenting with acute lower limb ischaemia caused by PAES.<sup>5,6</sup>

Acute limb ischaemia (ALI) in children is rare, but it may lead to limb loss or lifelong complications. Several well known factors, including embolic syndromes, thrombophilia, and hypercoagulable states,<sup>7</sup> can cause ALI. The diagnosis in children is often delayed since the first symptoms, including pain and claudication, are not reported by paediatric patients, hampering the possibility of early surgical treatments.

In this study, the aim was to report the experience of PAES in children and to analyse data from the literature in order to assess the severity of this disease and to determine

the factors characterising the diagnosis and outcome of treatment in paediatric patients.

**METHODS**

Four children (aged 8–16 years) were referred to hospital with ALI due to PAES between 2004 and 2009.

For the literature review, the following search strategy on the Medline database for popliteal entrapment syndrome was used: (“popliteal artery” [MeSH Terms] OR (“popliteal” [All Fields] AND “artery” [All Fields]) OR “popliteal artery” [All Fields]) AND entrapment [All Fields]. Altogether, 436 articles were identified. Literature research results were collected until March 2016. All papers that included patients under 18 years of age were selected for a detailed review (Fig. 1). Articles were excluded if the age of the treated patients was not mentioned in the paper or if the cases were not described. Three further articles were found from the reference lists of the selected papers. There were patients with PAES who were under 18 years old in 55

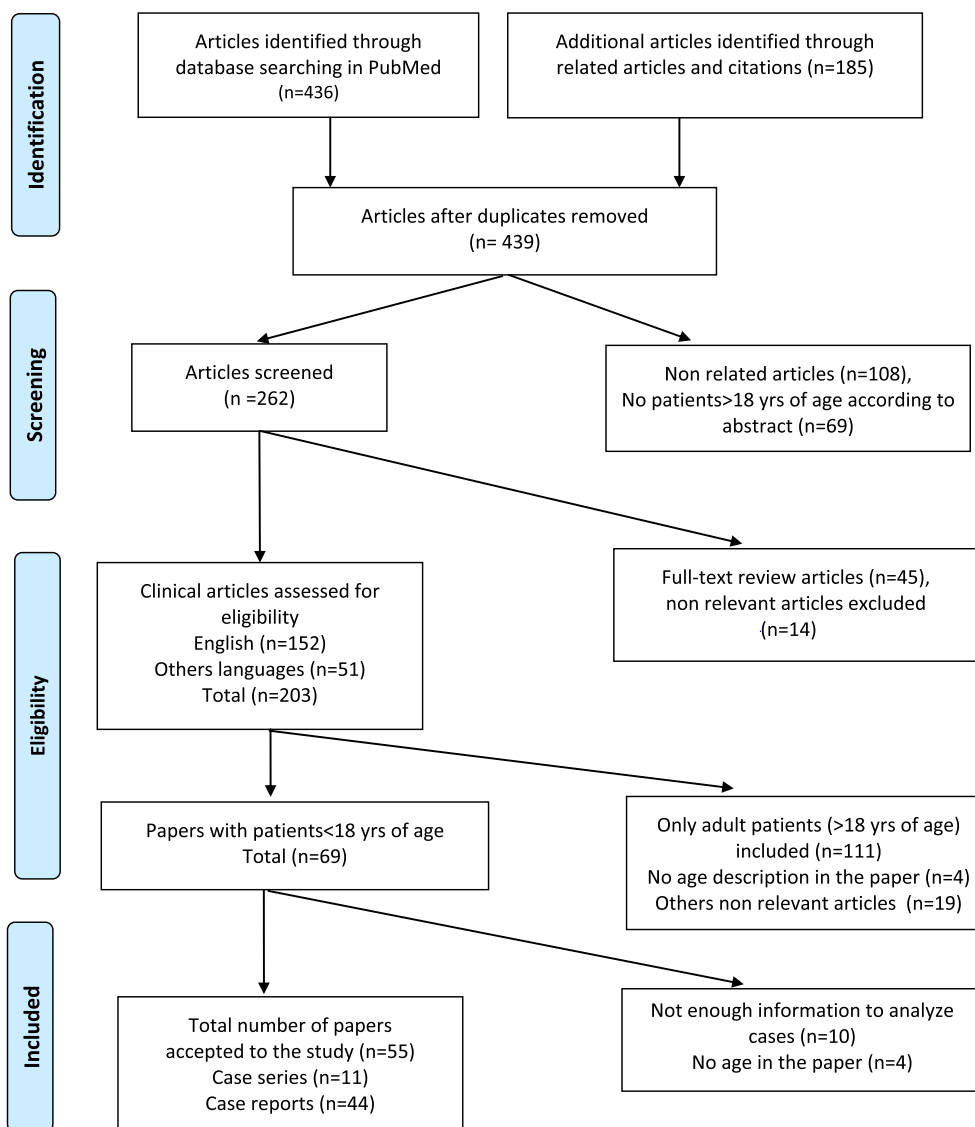


Figure 1. Strategy for the literature search.

papers. The PAES cases were classified according to the Love and Whelan classification modified by Rich<sup>8</sup> (Table 1). All 439 articles, including papers with no available abstract, were selected and analysed by two independent investigators. Papers in languages other than English were also analysed after translation. Data extraction was performed by the same two investigators and revised by a third one.

As shown in Appendix I, 79 cases were collected and analysed, including those from the authors' departments. It was decided to present the data for the overall group.

## RESULTS

### Own cases

During a 5 year period four cases of PAES were treated at the authors' institutions. Patients were 8–16 years of age and they all presented with acute symptoms. Popliteal and pedal pulses were absent and there was reduced motor and sensory function in all cases. Two patients had no claudication symptoms before the acute onset, the two others had claudication symptoms 10 days to a few weeks before the acute ischaemia. All patients had normal blood flow in the contralateral limb. Three patients underwent immediate duplex ultrasound (DUS) followed by computed tomography (CT) ( $n = 2$ ) or magnetic resonance angiography (MRA) ( $n = 1$ ) and an urgent operation thereafter. CT and MRA showed the diagnosis of PAES in all three cases. In one case the diagnosis was made on the operating table by intra-operative angiography: popliteal entrapment was evident during dorsiflexion of the foot. All patients underwent thrombectomy from the posterior approach and thereafter correction of the popliteal entrapment (Figs. 2 and 3) One patient underwent popliteo-popliteal interposition bypass with autologous vein and simultaneous fasciotomy. All patients recovered without major amputation. Two patients required minor amputations. Later, the PAES of the contralateral limb was operated on in two patients. After follow-up of 5, 7, 9, and 10 years, all patients were asymptomatic with patent popliteal arteries on DUS.

The results of the treatment in four cases are reported in Table 2 and detailed descriptions of the clinical signs and

**Table 1.** Classification of the PAES according to Love and Whelan modified by Rich and Hughes.<sup>8</sup>

Type I:	The PA has an aberrant medial course around the MHG, which has a normal insertion above the femoral condyle
Type II:	The MHG is inserted more laterally on the distal femur, with medial displacement of PA
Type III:	An aberrant accessory slip of MHG slings around and surrounds the normally positioned PA
Type IV:	The PA is located deep in the popliteal fossa and entrapped by the popliteus muscle or fibrous bands
Type V:	Any form of entrapment that involves both PA and PV.

PAES = popliteal artery entrapment syndrome; MHG = medial head of gastrocnemius; PA = popliteal artery; PV = popliteal vein.

symptoms, according to the Rutherford classification,<sup>9</sup> as well as the pathway of treatment are reported in Appendix II.

## RESULTS

Figure 1 summarises the strategy for literature research.

Table 3 summarises the clinical features of the paediatric patients with PAES reported, including the four children described herein.

Of 79 children, 54 (68%) were male. The mean age of onset was 15 years (range 7–17 years). In 27 (34%), the PAES was bilateral.

The most common symptoms on admission were claudication in 68 cases (64.2%) and ALI in 17 cases (18%); one patient presented with critical limb ischaemia (0.9%). In four cases (4%), symptoms were not reported.

In 14 (13%) of 27 (34%) children, an asymptomatic popliteal entrapment was also found in the contralateral limb. Among the children with bilateral PAES, the type of PAES was different in each leg in four cases (15%).

Type I PAES was present in 41 (39%), Type II in 23 (22%), Type III in 24 (23%), Type IV in 12 (11%), and Type V in two (2%) limbs. A functional PAES was present in one patient bilaterally. In two cases, the type of PAES was not reported.

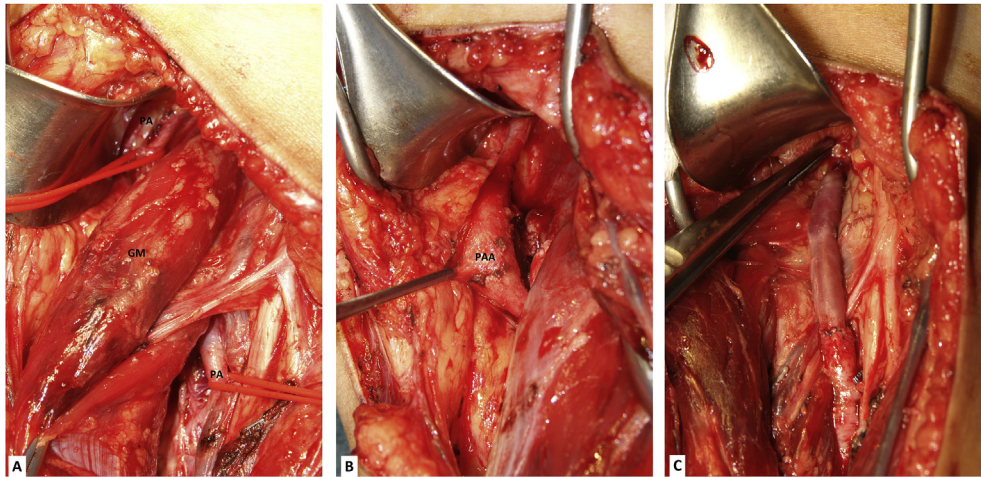
The treatment was surgical in the majority of cases ( $n = 96$ , 90.6%). In 41 cases (38.7%), only section of the musculo-tendinous structures was necessary. In the remaining 60 cases (56.6%), revascularisation was also required. In one patient, medical treatment was performed; in three the treatment was not reported,<sup>10,11</sup> and one patient declined any surgery.<sup>12</sup>

In 11 limbs (10%), the PAES was associated with a post-stenotic aneurysm, which was resected in all cases; a post-stenotic dilatation was observed in four cases (4%) without treatment. Complications occurred in four children (4%), but in all of the cases the patients obtained full recovery during long-term follow-up.

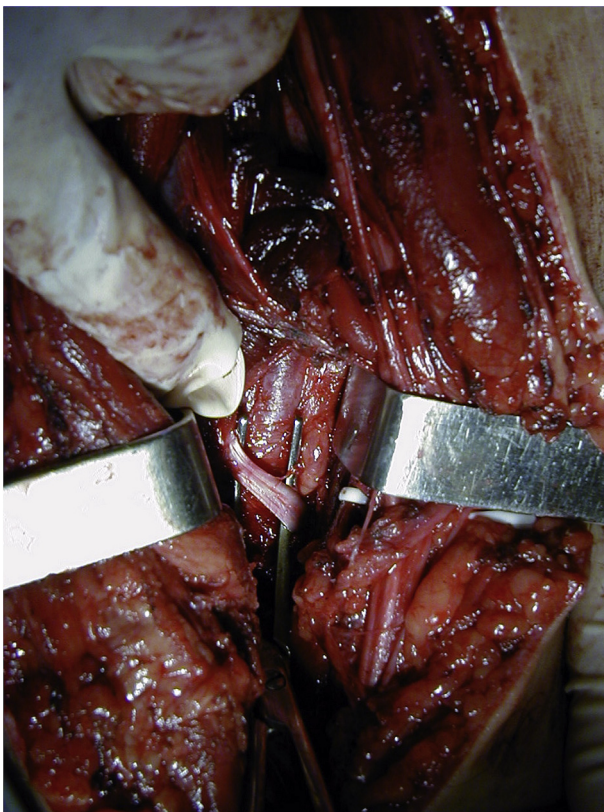
No major amputations were described. Only one patient (Case 3) had a transmetatarsal amputation and aponeurotomy, and in one patient four toes were partially amputated in a subsequent operation.<sup>13</sup>

## DISCUSSION

During the past 30 years, information on PAES has become increasingly available. However publications on PAES in children are scarce. Only a few isolated cases of PAES in children have been described, with rare reports of acute symptoms in this cohort of patients.<sup>3</sup> This is the largest report of PAES in children including the four reported here and 79 cases from the comprehensive review of the literature. The most common symptom of PAES in children was claudication but ALI was also very common. When the correct diagnosis is made and surgery carried out accordingly, the long-term results seem to be good as no patients underwent major amputation. However, it seems that the diagnosis is difficult in the non-acute phase, and because of this the diagnosis and operation is carried out in the acute



**Figure 2.** PAES in an 8 year old boy (Case 1). (A) The medial head of the gastrocnemius muscle (GM) is inserted more laterally on the distal femur, with medial displacement of the popliteal artery (PA) (type II). (B) Aneurysm of the popliteal artery (PAA). (C) Venous reconstruction after resection of the popliteal aneurysm.



**Figure 3.** A 16 year old boy (Case 4). The popliteal artery was located deep in the popliteal fossa and entrapped by a fibrous band.

situation in many cases and more extensive surgery is needed than when diagnosed and treated early.

Intermittent claudication in young patients is an unusual symptom, and the aetiology should be evaluated without delay. The cause is usually different from that typically seen in older patients suffering from an atherosclerotic condition.<sup>14</sup> Early diagnosis is of the utmost importance because PAES is a progressive condition and early treatment may

prevent serious complications,<sup>15,16</sup> and possibly, the need for amputation.<sup>17</sup> In fact, those patients who had ignored the initial symptoms of claudication and presented late frequently suffered from acute ischaemia caused by thrombosis of the diseased segment, complicated by distal embolisation with a poor distal runoff, thereby compromising a successful outcome.

The management of ALI in paediatric cases is difficult, since in contrast to adults, medical treatment with anti-coagulation is frequently applied as the first line therapy. However, such treatment may be unsuccessful for patients with PAES and only serves to delay a necessary surgical intervention. The other possible causes of ALI in young patients, such as premature accelerated atherosclerosis, adventitial cystic disease, microemboli, Takayasu's arteritis, collagen vascular disease, and coagulopathy,<sup>4,14</sup> should be excluded. All the authors' cases and 18% of the 79 cases found in the literature had ALI. In a recent meta-analysis on PAES including mostly adult patients, the median proportion of ALI was 11%.<sup>3</sup> Most of the patients with ALI in the current report of 79 patients, described a period of claudication (ranging from a few days to a few years) with worsening pain while walking and running. If such patients sought medical attention belatedly, acute ischaemia had frequently already developed, often entailing a limb threatening prognosis. When an occlusion of the popliteal artery exists in a young patient, the possibility of PAES should be suspected. Misdiagnosed PAES can induce surgeons to apply inappropriate techniques, and even stenting of these lesions has been described.<sup>15</sup>

DUS is a non-invasive and effective technique for the early diagnosis of PAES and may even be the key investigation when the popliteal artery is still patent. A decrease in the peak systolic flow of the popliteal artery could be indicative of PAES, with provocative manoeuvres obtained with the contraction of the calf muscles.<sup>18,19</sup> In the earlier reports, arteriography was used as a diagnostic and sometimes as a therapeutic technique.<sup>20</sup> Recently, this invasive

**Table 2.** Four cases reported in the article.

Patient	Sex	Age	Side	Symptoms	Delay	Examination	Type of PAES	Popliteal artery lesions	Treatment	Outcome
1	M	8	R	ALI	2 d	DUS + CT scan	II	Aneurysm	Thromb + PP	8 y OK
			L	Asymptom	—	DUS + CT scan	IV	Normal	Myo	8 y OK
2	F	13	L	ALI toe necrosis	3 mo	DUS + MRI	III	Thrombosis	Thromb + Myo	9 y OK
			R	IC	—		III	Compression	Myo	9 y OK
3	M	8	L	ALI	5 h	Angiography	IV	Thrombosis	T + PP + Apon + Trans-Met	7 y OK
4	M	16	L	ALI	3 wk		IV	Thrombosis	T + Myo	10 y OK

ALI = acute limb ischemia; Apon = aponeurotomy; BP = bypass; DUS: Doppler ultrasonography; F = female; IC = intermittent claudication; L = left; M = male; mo = month(s); Myo = myotomy; PP = popliteo-popliteal bypass; R = right; T = thrombectomy; Trans-Met = trans-metatarsal amputation; wk = week(s); Y = year(s).

**Table 3.** Analysis of PAES in 79 children. Results of clinical features, type of PAES, treatment and outcome.

	Number %	
No. of cases	79	
Age, years	15	Range (7–17)
Male	54	68
Female	19	24
NR sex	6	8
Total limbs	106	
Bilateral	27	34
Claudication	68	64
Acute ischaemia	19	18
Critical limb ischaemia	1	1
Asymptomatic	14	13
NR symptoms	4	4
Type of PAES		
I	41	39
II	23	22
III	24	23
IV	12	11
V	2	2
Functional PAES	2	2
NR type	2	2
Surgery	95	90
No surgery	4	4
NR surgery	3	3
Myotomy	40	38
Revascularisations	58	55
Surgical revasc.	52	87
Endovascular revasc.	6	10
Aneurysm or dilatation	15	14
Minor amputations	2	2
Outcome 1–13, 3 years (complications)	4	4

NR = not reported; PAES = popliteal artery entrapment syndrome.

technique has been replaced by non-invasive techniques, including in addition to DUS, CTA, MRI, and MRA.<sup>11,18</sup>

The main surgical treatment is releasing the popliteal entrapment, establishing a normal anatomy, and restoring a normal arterial flow.<sup>21–25</sup> Vascular reconstruction with venous bypass is the most frequent procedure in the literature.<sup>26,27</sup> Some authors suggest endovascular treatment combined with thrombolysis to revascularise the limb, followed by surgical decompression.<sup>13,21</sup> Thrombolysis may be useful in cases of ALI with distal embolism to restore the distal outflow.<sup>22</sup> Few studies reported a long-term follow-up with a satisfactory outcome 5 years after the surgical

treatment (84–92%).<sup>28</sup> When musculo-tendinous section was done alone, the long term patency was better than when vascular reconstruction was needed.<sup>29</sup> Furthermore, a short interposition vein graft had better patency than cases where a long bypass vein graft was needed.<sup>30</sup>

The reported incidence of bilateral involvement varies in the literature, with a median prevalence of 38.25%.<sup>3</sup> When an anatomical anomaly exists, even if asymptomatic, surgical repair is suggested to prevent irreversible arterial damage.<sup>6</sup> As reported in the review of the literature, children with bilateral PAES had myotomy combined with vascular reconstruction in the first operated limb. In 14 patients (13%), the contralateral limb was asymptomatic and most of them ( $n = 10$ ) underwent surgical reconstruction because of a positive response to provocative manoeuvres.

Compression by musculo-tendinous structures causes microtrauma to the artery, which may lead in the long term to aneurysm formation in the popliteal artery<sup>16,24</sup> and, at times, thrombosis of the collateral circulation, causing acute ischaemia. In this literature review, a post-stenotic aneurysm was found in eight limbs, frequently associated with distal embolisation. When arterial wall damage has occurred with an aneurysm, stenosis, or occlusion, surgical revascularisation with a venous bypass is indicated. A significant number of authors recommend the use of an autologous vein as a first option for reconstruction.<sup>27</sup>

Although PAES is uncommon and difficult to diagnose clinically, it must be considered in children who present with ALI or claudication and no cardiovascular risk factors exist. A careful anamnesis should investigate all possible early signs of PAES. In children complaining of even minor symptoms, PAES should be suspected and duplex scanning, combined with CT and MRI, should be performed.

If the imaging and the treatment methods are basically the same in both the adult and paediatric populations, the diagnosis is the key issue in the management of PAES as far as both populations is concerned. In the adult population, the diagnosis of the PAES is mainly raised before specific symptoms that lead to adequate imaging and then to diagnosis. Compared with the adult population, the diagnosis of PAES in children may be more challenging because their complaints may be incorrectly described or misunderstood by those closest to them. This may lead to delayed

diagnosis and more frequent intervention in the emergency setting.

Following the results of the present study, the place of conservative management of PAES in the paediatric population is limited because it may lead to irreversible deterioration of the arterial outflow with the risk of limb loss. A simple correction of the anatomical variation, meaning sectioning the musculo-tendinous structures, may be an adequate therapy in children with no arterial disease. In other cases, the division of any entrapping structures, combined with an arterial reconstruction with an autologous vein and possibly aneurysm resection, is recommended. In the case of acute ischaemia, immediate DUS confirms occlusion of the artery and anatomy of the popliteal artery and adjacent structures can be seen on CT or MRA. However, if immediate CT/MRA is not available, the operation has to take place without more detailed imaging of the popliteal structures. In these cases three steps can lead to a correct diagnosis: anamnesis with history of previous claudication, DUS of the contralateral limb with provocative manoeuvres, and, finally, a post-thrombectomy angiogram in the operating room with dynamic manoeuvres. Low molecular weight heparin or heparin infusion should be started at the time of diagnosis and because the ischaemia is most frequently severe, urgent revascularisation is needed. The type of revascularisation depends on the arterial damage, but in acute cases arterial reconstruction with autologous vein is usually needed. If arterial grafting is necessary, autologous great saphenous vein is the material of choice. This graft can be harvested at mid thigh level even in the prone position.

The external compression of the popliteal artery must be taken care of as in elective cases. If any doubt about the result of thrombectomy exists, a completion angiogram is recommended. In cases of clear PAES in the imaging studies and at operation, no other studies on the etiology of ALI are needed. For bilateral PAES it is recommended that both sides are treated, although asymptomatic, to avoid later acute ischaemia and increased risk of limb loss.

## CONCLUSIONS

A PAES diagnosis can be difficult, since children may ignore the symptoms. As a result, the diagnosis is often delayed. ALI in paediatric patients is rare but may lead to limb loss and lifelong complications. Symptomatic PAES in children must be considered a severe condition needing urgent investigation in order to avoid any delays in treatment. The long-term outcome after surgical treatment, including correction of the anatomical anomaly as well as vascular reconstruction, is generally satisfactory and carries a low complication rate.

## CONFLICT OF INTEREST

None.

## FUNDING

None.

## APPENDIX A. SUPPLEMENTARY DATA

Supplementary data related to this article can be found at <http://dx.doi.org/10.1016/j.ejvs.2016.12.032>.

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