Ruptured popliteal artery aneurysm

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Background: Popliteal artery aneurysms (PAAs) are generally complicated by thrombosis and distal embolization, whereas rupture is rare. The aim of this study was to describe the clinical characteristics and outcome in a cohort of patients who had surgery for ruptured PAA (rPAA).

Methods: Operations for rPAA were identified from the Swedish Vascular Registry, Swedvasc, 1987–2012. Medical records and imaging were reviewed. Comparison was made with patients treated for PAA without rupture.

Results: Forty-five patients with rPAA were identified. The proportion with rupture among those operated on for PAA was 2.5 per cent. Patients with rPAA were 8 years older (77.7 *versus* 69.7 years; P < 0.001), had more lung and heart disease (P = 0.003 and P = 0.019 respectively), and a larger mean popliteal aneurysm diameter (63.7 *versus* 30.9 mm; P < 0.001) than patients with PAA treated for other indications. At time of surgery, 22 of 45 patients were already receiving anticoagulants, seven for concomitant deep venous thrombosis (DVT) in the affected leg. There was extensive swelling of the whole leg in 20 patients. In 27 patients, the initial diagnosis was DVT or a Baker's cyst. All patients underwent surgery, all but three by the open method. There were four amputations, all performed within 1 week of surgery. One year after surgery, 26 of the 45 patients were alive. Among these, the reconstructions were patent in 20 of 22 patients.

Conclusion: The diagnosis of rPAA is difficult, and often delayed. The condition affects old patients, who often are on anticoagulation treatment and have large aneurysms. The immediate surgical results are acceptable, but the condition is associated with a high risk of death within the first year after surgery.

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Introduction

Popliteal artery aneurysm (PAA) is the most common of the peripheral aneurysms, but is still rare. Trickett and colleagues¹ screened 1070 men aged 65-80 years, identifying 11 (1.0 per cent) with a PAA diameter between 15 and 26 mm¹. Contemporary population-based prevalence data are lacking. The fact that the prevalence of abdominal aortic aneurysm (AAA) is decreasing^{2,3} suggests that the prevalence of PAA could also be falling^{4,5}, as PAA is often associated with multianeurysm disease. The number of operations for PAA in Sweden has doubled from 1994–2001⁶ to 2008–2012⁷, although this may be a result of improved diagnosis.

Thrombosis and distal embolization are the most common complications of PAA, and can lead to severe limb-threatening ischaemia^{6,8}. The proportion of ruptured

popliteal aneurysms (rPAAs), among all patients operated on for PAA, is 2–5 per cent in larger series^{6,7,9,10}. Many centres use a diameter of 2 cm, with or without thrombus, as the indication for prophylactic surgery, supported by the American College of Cardiology/American Heart Association peripheral artery disease 2005 guidelines¹¹. The risk of rupture of PAA with different diameters is virtually unknown.

Previous publications on PAA consist of case reports and small case series. The condition is often misdiagnosed as deep venous thrombosis (DVT), ruptured Baker's cyst or acute limb ischaemia^{7,12,13}.

The aim of this study was to describe the clinical characteristics and outcome in patients with rPAA, and to compare them with those of patients who were treated for non-ruptured PAA.

Methods

The Swedish Vascular Registry, Swedvasc, was created in January 1987 and since 1992 has registered more than 90 per cent of all open and endovascular procedures¹⁴. In 2008, a special module for PAA was added to the registry. Independent international validation has shown excellent external and internal validity¹⁵. There has also been a specific validation of the registration of patients treated for PAA in the Swedvasc. Ravn and colleagues⁴ took advantage of the fact that PAAs often are bilateral. Of the 146 bilateral procedures, 141 (96.6 per cent) had been reported to the registry.

Two cohorts were combined for the present study. In 2007, Ravn *et al.*⁶ used data from Swedvasc to study a nationwide cohort of 717 legs treated for PAA, of which 24 ($3\cdot3$ per cent) were treated for rupture. In a recently published study⁷ using the same registry, 592 PAAs treated between 2008 and 2012 were described. Of these, 13 had been rPAAs ($2\cdot2$ per cent). Another 12 separate patients with rPAA during 2002–2008 were added, as well as one operated on in 2001 but registered in 2002. The patients treated for rPAA during these three time intervals were merged into one data set. Prospectively entered registry data were supplemented with data from the case records and imaging, collected retrospectively from 20 different hospitals.

In all, 50 patients treated for rPAA were identified but, when the case records were scrutinized, one was registered twice and four patients had been treated for post-traumatic pseudoaneurysms. Thus, 45 patients treated for rPAA remained for study.

Characteristics of the patients with rPAA were compared with those of patients with PAA treated for other indications, using original data from the two previously published nationwide studies^{6,7}. In the nationwide studies, there was some difference in the data collected. The earlier cohort had data on size and other aneurysm disease⁶. The latter had data on background disease⁷.

The Ethics Committee of the Uppsala region approved the study.

Statistical analysis

The distribution of continuous variables was assessed by histogram and Q-Q plots. Independent-samples t test was used to evaluate differences in continuous variables between groups, and Pearson correlation coefficient to estimate the correlation between patient age and rPAA size. Differences in proportions were evaluated using Fisher's exact test. All statistical analyses were performed using the software package SPSS version 23.0 (IBM, Armonk, New York, USA).

Results

To calculate the proportion of patients who had surgery for rPAA, the time intervals 1987-2002 and 2008-2012were used, for which complete data were available on the total number of repairs (1304) as well as the number of operations for rPAA (33); the proportion was 2.5 per cent.

Patients with rPAA were older (P < 0.001) and had more lung and heart disease (P = 0.003 and P = 0.019 respectively) than with those with PAA treated for other indications (*Table 1*). Among the patients with non-ruptured PAA, there were no significant differences in this regard between those treated urgently or electively. The maximum diameter of the rPAA was assessed in 38 patients, by CT (17), ultrasonography (16), MRI (2) and perioperative measurement (3) (*Fig. 1*). The mean maximum rPAA diameter was 63.7 (range 25–200) mm (*Table 2*). There was no correlation between patient age and rPAA diameter (Pearson r = 0.046, P = 0.777).

Treatment

Most patients had open surgery by a medial approach. The surgical techniques and outcomes are described in *Table 2*. Fasciotomy was performed in 12 of 45 patients and four legs were amputated within 30 days. At 1 year, 26 patients were alive. The cause of death was uncertain in most patients who died within 1 year after surgery, owing to a low autopsy rate, but three were known to have died from aortic rupture/dissection and two from cancer.

Twenty-two of 45 patients were being treated with anticoagulants at the time of surgery for rPAA, 19 with oral anticoagulants and three with low molecular weight heparins. Among these, seven had the treatment started within 2 months before being diagnosed with rPAA because of suspicion of DVT. The remaining 15 patients were receiving chronic anticoagulation treatment, mostly for atrial fibrillation or pulmonary embolism. Seven patients presented with critical leg ischaemia. One patient was hypotensive on admission, but quickly stabilized; another had fainted at home, but was stable on admission. No patient was in severe shock at presentation. Almost half of the patients (18 of 42) presented on the day their symptoms started and another 11 (in all 29 of 42) within 1 week. It was sometimes difficult to identify the exact date of the rupture, as most ruptures were contained, and the diagnosis was often delayed.

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 Table 1 Clinical characteristics of patients who had surgery for ruptured or non-ruptured popliteal artery aneurysm

	Rupture (<i>n</i> = 45)	Other indications $(n = 579 \dagger \text{ or } 693 \ddagger)$	P§
Mean age (years)*	77.7 (59–96)	69.7 (42-102)†	< 0.001 ¶
Male sex	43 of 45 (96)	554 of 579 (95·7)†	1.000
Heart disease	18 of 44 (41)	131 of 542 (24·2)†	0.019
Hypertension	22 of 43 (51)	365 of 544 (67·1)†	0.063
Respiratory disease	12 of 43 (28)	60 of 529 (11·3)†	0.003
Diabetes	3 of 44 (7)	77 of 574 (13·4)†	0.250
Cerebrovascular disease	9 of 43 (21)	59 of 537 (11·0)†	0.079
Smoker			
Never	6 of 25 (24)	89 of 440 (20·2)†	0.686
Former	14 of 25 (56)	229 of 440 (52·0)†	
Active	5 of 25 (20)	122 of 440 (27·7)†	
PAA diameter (mm)*	63·7 (25−200) (<i>n</i> = 38)	30·9 (10−150) (<i>n</i> = 513)‡	< 0.001¶
Concomitant AAA	14 of 39 (36)	236 of 687 (34·4)‡	0.863
Bilateral PAA	19 of 37 (51)	388 of 687 (56·5)‡	0.611

Values in parentheses are percentages unless indicated otherwise: *values are mean (range). The clinical subgroups refer to legs, not patients. †Original data from 579 patients with non-ruptured popliteal artery aneurysm (PAA) treated between 2008 and 2012; ‡original data from 693 legs with non-ruptured PAA treated between 1987 and 2002. AAA, abdominal aortic aneurysm. §Fisher's exact test, except ¶independent-samples *t* test.

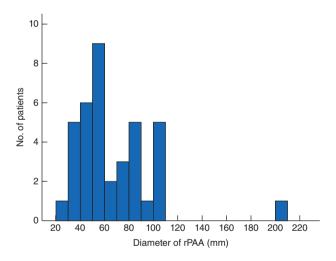


Fig. 1 Ruptured popliteal artery (rPAA) diameter

The initial diagnosis was rPAA in only eight of the 45 patients, half of whom were known to have a PAA. Twenty-seven patients had a preliminary diagnosis of DVT or a Baker's cyst. Bleeding because of minor trauma or anticoagulation was suspected in seven patients and no preliminary diagnosis could be identified in the remaining three. All patients had swelling, and in 20 the whole leg was affected. Two patients, with known PAAs, had sepsis; their aneurysms became infected and ruptured.

 Table 2
 Surgical technique and outcome in 45 patients treated

 for ruptured popliteal aneurysm

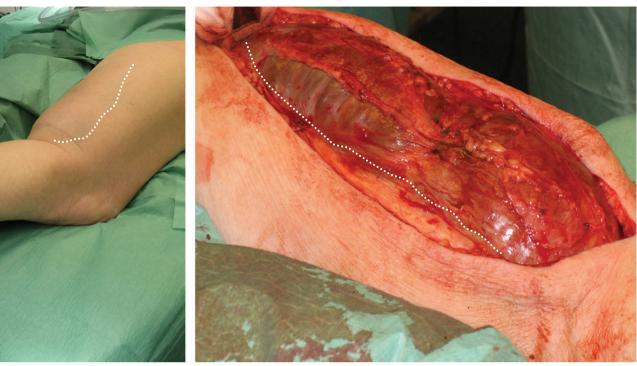
	No of patients ($n = 45$)
Surgical approach	
Endovascular	3
Open, medial	35
Open, posterior	3
Open, ligation only	3
Open, direct anastomosis	1
Graft material ($n = 38$)	
Venous	22
Prosthetic	14
Composite	2
Outflow vessels $(n = 33)$	
0	5
1	12
2	4
3	12
Fasciotomy	12
30-day outcomes	
Patency	35 of 39
Amputation	4 of 45
Survival	40 of 45
1-year outcomes	
Patency	20 of 22
Amputation	4 of 44*
Survival	26 of 45

*The total number of amputations did not increase but one of the patients alive at 1 year was lost to follow-up.

Discussion

Patients with rPAA were 8 years older than those treated for non-ruptured PAA. The aneurysms were larger and the initial clinical picture misleading, resulting in frequent delays in diagnosis. The immediate outcome after surgery was good; however, there was a high risk of death within 1 year, not just explained by old age and frequent cardiopulmonary co-morbidities. There were at least three cases of fatal aortic rupture/dissection. CT angiography of the aorta should be considered in patients who present with rPAA.

Rupture of PAA is an uncommon event in an uncommon disease. In other aneurysm disease, the major concern is the risk of rupture, which also guides the indication for treatment. In patients with PAA, however, the most common complication is ischaemia, and little is known about the few aneurysms that rupture. The largest cohorts before this investigation were published in 1953¹⁶ and 1962¹⁷, including 16 and 11 patients respectively. In neither of these studies were the specific characteristics of patients with rPAAs described, and pseudoaneurysms after trauma were also included. Roggo and co-workers¹⁰ reported on 252 patients treated for PAA, of which six (2·4 per cent) were ruptured. Sie and colleagues⁹ analysed 89 patients who had surgery for 124 PAAs, and the six with rPAAs (4·8 per cent) were described in detail. The main finding



a Lateral circumference

b Medial circumference

Fig. 2 Ruptured popliteal artery aneurysm in the right leg. Note the bluish discoloration on top of the bulge just above the hollow of the knee. The dotted line delineates the a lateral and b medial circumference of the haematoma (courtesy of S. Acosta, Vascular Centre Malmö, Malmö, Sweden, and S. Mathiesen, Hamar Hospital, Hamar, Norway)

was that the clinical presentation varied, with different and often incorrect preliminary diagnoses, a finding verified in the present study.

Here, the mean rPAA diameter of 63.7 mm was twice that of PAAs in patients who had surgery for other indications. It was tricky to measure the diameter of some of the largest rPAAs owing to difficulty in defining the wall of the aneurysm in a large haematoma. PAAs may reach a diameter over 10 cm without rupturing, and yet six ruptured in the present series despite having a diameter of only 30-40 mm. It is known that even small-diameter PAAs sometimes rupture, and the risk was estimated recently among patients with AAAs and internal iliac artery aneurysms (IIAAs). In Finland, 8 per cent of ruptured AAAs were below 55 mm in diameter¹⁸. In a multinational study¹⁹, 6 per cent of IIAAs were smaller than 40 mm at the time of rupture. Thus, factors other than size influence the risk of rupture. In the present study, a known PAA ruptured during treatment for sepsis in two patients, suggesting that the aneurysms could have been infected, resulting in rapid expansion and rupture. The findings during surgery in both patients, and positive culture in one, confirmed the diagnosis of a mycotic aneurysm. Infection has been associated with a high risk of rupture of AAA^{20,21}.

Almost half of the patients were on an anticoagulant at the time of rupture. This was an unexpected and novel finding. Most had been treated for a long time and the anticoagulation may explain why the aneurysms grew so large without thrombosis or embolization. In others, swelling of the leg associated with a contained rupture may have been misinterpreted as a DVT. This was suspected in four of the seven patients who had anticoagulation treatment initiated within 2 months of the diagnosis and treatment of rPAA.

PAAs usually rupture into the popliteal space, confined by muscles and tendons. The main symptoms are pain and swelling, and the patients were often evaluated initially by specialists in internal medicine or orthopaedics. Twenty-seven of the 45 patients in the present study had a preliminary diagnosis of DVT or Baker's cyst. Compression of the popliteal vein²² and development of DVT²³ are well known complications of large PAAs, but 20 of the patients with an rPAA presented with swelling of the entire leg, which should not be expected when a DVT develops secondary to compression from a PAA. Pressure from a large PAA, or the haematoma in the event of rupture, can also cause neurological pain^{24,25}, adding further complexity to the challenge of reaching the correct diagnosis (*Fig. 2*).

The results of surgery were acceptable, given the challenge of this rare condition. Among those who survived 1 year, all but two reconstructions were patent. Of four patients who needed an amputation, three presented with both ischaemia and rupture.

This study has limitations. The small numbers of rPAAs make the investigation prone to type II statistical error. However, it was possible to identify several differences between the small cohort of patients with rPAA and the larger cohort treated for non-ruptured PAA. Furthermore, a long time interval was studied. The findings might have been different if a modern patient cohort alone had been investigated. Imaging and clinical pathways may have improved over time, although no such improvement could be detected in this small cohort.

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