

General Reviews/Meta-analysis

Spontaneous Ilio-Iliac Arteriovenous Fistula from Rupture of an Iliac Aneurysm: A Systematic Review

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Background: Spontaneous arteriovenous fistulas (AVF) caused by iliac aneurysms are a rare condition with possible dramatic complications due to secondary hemodynamic changes. Diagnosis can be challenging because patients may present with progressive cardiac failure or even hemodynamic shock as primary symptom. Due to the rarity of the condition, data are scarce and treatment decisions are challenging. The aim of this systematic review is to give an overview of the symptoms, treatment possibilities, and patient outcomes.

Methods: Literature searches were performed in PubMed, Embase, Web of Science, and Scopus. Case reports and literature reviews were included in the review. The literature review was performed by 2 independent reviewers according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. A third reviewer was available in case of disagreement. The study was registered in Prospero (ID CRD42022335318). All cases of isolated, iliac atherosclerotic aneurysms with spontaneous fistulization into an iliac vein were included.

Results: Fifty articles were included, resulting in 62 cases. A case from our own center was included, bringing the total up to 63 cases. Median age was 71 years, ranging from 41 to 87 years. 87.3% of patients were male, 6.3% were female, and in 6.3% sex was not reported. The duration of symptoms until presentation ranged from less than an hour to 6 years. 73.0% was treated with open surgery and 17.5% was treated by endovascular way, with 4 reinterventions in the endovascular group. There was an overall mortality rate of 9.5%.

Conclusions: Although rare, iliac AVF might cause acute therapy-resistant heart failure and hemodynamic instability. In patients with acute heart failure, especially when combined with a pulsating mass with accompanying bruit or thrill and unilateral swollen leg, an AVF should be suspected. Surgical treatment of AVF has an excellent outcome, provided that the condition had been diagnosed preoperatively.

INTRODUCTION

Acquired iliac arteriovenous fistulas (AVF) are a rare condition with several possible causes. According to literature, up to 84% of iliac AVF are caused by trauma, including procedure-related (iatrogenic), while the remaining 16% arise spontaneously, usually originating from fistulization of an iliac aneurysm.^{1–3} Ilio-iliac AVF only account for 1.8% to 4% of major abdominal AVF.^{4,5} An abruptly emerged iliac AVF can present with hemodynamic instability or cardiac failure and pose a diagnostic challenge.^{6–8} Several cases have been reported since the first one by Zajtchuk et al. in 1971,⁹ but

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Fig. 1. Patient CT angiography shows rapid enhancement of the vena cava during arterial phase.

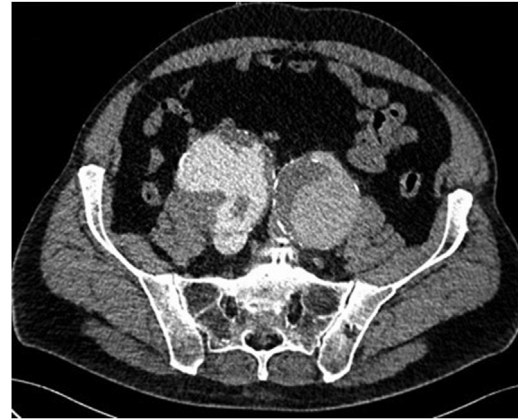


Fig. 2. Patient CT angiography shows bilateral aneurysms of the common iliac arteries. Despite the suspicion of rupture, no retroperitoneal hematoma is present.

it remains an unknown condition. In 1986, McAuley et al. described a triad of symptoms accompanying spontaneous iliac AVF: 1/a pulsatile abdominal mass with accompanying bruit or thrill, 2/high-output cardiac failure (either progressively or fulminant onset), and 3/unilateral lower limb ischemia or venous engorgement.¹ Based on a case of our own center, we performed a systematic review of the literature and enlisted the possible symptoms, consequences, and treatment options for spontaneous ilio-iliac AVF.

Case Description

A 67-year-old man consulted in another hospital because of severe abdominal pain with sudden onset the day before. The pain radiated to his back and had increased overnight. Clinical examination revealed a painful abdomen, absent femoral pulses, and a systolic blood pressure of 60 mm Hg with immeasurable diastolic blood pressure. Hemoglobin level was 6.77 g/dL (reference 12.9–16.9 g/dL). A contrast-enhanced computed tomography (CT) scan showed bilateral iliac aneurysms, raising the suspicion of a ruptured aneurysm. The patient was immediately transferred to the operating room. After inflation of a Resuscitative Endovascular Balloon Occlusion of the Aorta (REBOA) balloon to occlude the infrarenal aorta, an angiography of the right iliac arteries to evaluate the presence of landing zones and to confirm the diagnosis revealed a massive AVF between the aneurysmatic common iliac artery and the common iliac vein.

Considering the expected difficulty of open repair, the patient was transferred to our tertiary center with the REBOA balloon in situ. Review of the CT scan revealed bilateral common iliac

aneurysms without retroperitoneal hematoma and equal contrast density in the aorta and inferior vena cava suggesting a massive AVF at the level of the iliac vessels (Figs. 1 and 2). Open surgery was preferred over endovascular repair, as both internal iliac arteries (IIAs) were included in the aneurysms. A median laparotomy was performed with clamping of the infrarenal aorta and resection of both iliac aneurysms. The laceration in the right common iliac vein caused by the aneurysm rupture was closed and an infrarenal aorto-femoral bifurcation graft was used for reconstruction. Due to the REBOA occlusion time of more than 6 hr, thrombosis of both legs had occurred necessitating thrombectomy and bilateral 4 compartment fasciotomies. Because of long-lasting hemodynamic instability due to the duration of the transfer and surgery, the patient developed a refractory shock resulting in multiple organ failure. The patient died within 12 hr postoperatively.

MATERIALS AND METHODS

A literature search was performed in the databases PubMed, Embase, Scopus, and Web of Science according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines (Fig. 3). Two independent reviewers (G.S. and C.A.) performed the complete literature search using the same search strings. There were no conflicts, so the third reviewer (R.C.) was not addressed. For PubMed, the MeSH terms “Arteriovenous Fistula” and “Iliac Aneurysm” were combined. For Embase, a simple keyword search using ‘arteriovenous fistula’ and ‘iliac artery aneurysm’ was performed. In Scopus, a search string was used with the terms ‘arteriovenous

Table I. Overview of included cases

Case number	Author	Publication year	Sex	Age	Duration of symptoms	Artery	Vein	Technique	Treatment	Postop course	Survival
1	Gedeon et al. ^{3*}	1970				RCIA	RCIV	open	Interposition graft		yes
2	Zajtchuk et al. ⁹	1971	M	69	5 days	RCIA	RCIV	open	Direct closure fistula, bilateral ligation IIA, aorto-iliac bifurcation graft	uneventful, resolution of symptoms	yes
3	Birnholz ¹⁰	1973	M	82	1 year	LCIA	LCIV	open	Aorto-iliac bifurcation graft		yes
4	Hines et al. ¹¹	1978	M	60	7 weeks	LCIA	LCIV	open	Direct closure fistula, iliac artery graft, oversewing IIA	uneventful, resolution of symptoms	yes
5	Rubio et al. ¹²	1978	M	59	11 days	RCIA	RCIV	open	Direct closure fistula, aorto-femoral bifurcation graft	uneventful	yes
6	Datta and Vickery ^{1†}	1979	M	66	2 weeks	LCIA	LCIV	open	Aorto-iliac bifurcation graft		yes
7	Bacourt et al. ^{3*}	1980				LCIA	LCIV	open	Interposition graft		yes
8	Fourmestraux et al. ^{1†}	1980	M	71	1 day	RCIA	RCIV	open	Unilateral aorto-iliac graft		yes
9	Lo et al. ^{1†}	1981	M	74	1 day	RCIA	LCIV	open	Unilateral aorto-femoral graft		yes
10	Wills et al. ¹³	1982	M	60	10 days	LCIA	LCIV	open	Ligation aneurysms and LIV, aorto-femoral bifurcation graft	uneventful, resolution of symptoms	yes
11	DuToit et al. ^{1†}	1983	M	63	6 years	LCIA	LCIV	open	Aorto-iliac bifurcation graft		yes
12	Richards, D. ¹⁴	1984	M	80	3 hr	RCIA	RCIV	Open	Ligation RCIA, REIA and RIIA, bypass LEIA to REIA	uneventful, resolution of symptoms	yes
13	Duda et al. ^{1†}	1984	M	60	1 day	RCIA	RCIV	Open	Interposition graft		yes
14	Bharadwaj et al. ¹⁵	1985	M	63	2 months	LCIA	LCIV	Open	Ligation fistula and aneurysms, aorto-iliac bifurcation graft.	uneventful, resolution of symptoms	yes
15	Campbell et al. ¹⁶	1985	F	76	3 weeks	RIIA	RCIV	Open	Direct closure fistula, iliac interposition graft	persistent swelling gradually decreasing	yes
16	Duppler et al. ^{3*}	1985				RCIA	RCIV	Open	interposition graft		yes
17	McAuley et al. ¹	1986	M	74	Acute	RCIA	RCIV	Open	Direct closure fistula, aorto-iliac bifurcation graft	perioperative myocardial infarction	yes
18	Falk et al. ¹⁷	1986	M		10 days	LCIA	LCIV	Open	Direct closure fistula, aorto-iliac bifurcation graft	uneventful, resolution of symptoms	yes
19	Weimann et al. ³	1987	M	71	Acute	LCIA	LCIV	open	Direct closure fistula, iliac interposition graft.	uneventful, resolution of symptoms	yes
20	Cabot et al. ¹⁸	1988	M	72	1 month	RCIA	RCIV	open	Direct closure fistula, aorto-iliac bifurcation graft	temporary impairment of liver function	yes
21	Odagiri et al. ¹⁹	1988	M	59	1 year	LCIA	LCIV	open			

22	Redmond et al. ²⁰	1988	M	69		IIIA	IIIV	open	Direct closure fistula, aorto-iliac bifurcation graft	uneventful, resolutions of symptoms	yes
23	Walstra et al. ²¹	1989	M	84	4 weeks	RCIA	RCIV	open	Ligation RIIA, direct closure fistula, iliac interposition graft	uneventful, resolution of symptoms	yes
24	Turse et al. ⁷	1989	M	71	1 week	LCIA	LCIV	open	Direct closure fistula, aorto-iliac bifurcation graft	thrombophlebitis left leg, resolution with routine measures. Resolution of symptoms	yes
25	Brewster et al. ² - case 11	1991	M	62		RCIA		open	Direct closure fistula, iliac interposition graft		yes
26	Brewster et al. ² - case 12	1991	M	56		LCIA		open	Direct closure fistula, aorto-iliac bifurcation graft		yes
27	Brewster et al. ² - case 13	1991	M	79		IIIA		open	Direct closure fistula, aorto-iliac bifurcation graft.		yes
28	Brewster et al. ² - case 14	1991	M	64		RCIA		open	Direct closure fistula, iliac interposition graft		yes
29	Gilling-Smith et al. ²²	1991	M	66	3 years	RCIA	both CIV	open	Direct closure fistula, iliac interposition graft	progressive multisystem failure and coagulopathy	no
30	Flarup et al. ²³ - case 1	1999	M	69	6 hr	RCIA	RCIV	open	Direct closure fistula, aorto-iliac bifurcation graft.		no
31	Flarup, S. ²³ - case 2	1999	M	68	1 month	LCIA	LCIV	open	Aorto-iliac bifurcation graft	uneventful, resolution of symptoms	yes
32	Glovicski et al. ⁵³	1999	M	73	10 days	LCIA	LCIV	open	Direct closure fistula, LIIA oversewn, aorto-iliac bifurcation graft	uneventful, resolution of symptoms	yes
33	Vallina et al. ²⁴	2000	M	74	4 days	RIA	RIV	open	Direct closure fistula, aorto-iliac bifurcation graft	mechanical breathing and inotropy—resolution of symptoms	yes
34	Chiorean et al. ²⁵	2001	M	73	2 weeks	LCIA	LCIV	unknown		uneventful, resolution of symptoms	yes
35	Char et al. ²⁶	2003	M	73		RIIA	RCIV	endovascular	See Table IV	uneventful, resolution of symptoms	yes
36	Krishna et al. ²⁷	2005	M	73	3 weeks	RCIA	LCIV	open	Aorto-iliac bifurcation graft		yes
37	Tan et al. ²⁸	2006	F	81	5 days	RIIA	RIIV	failed attempt at coiling of the fistula	Conservative treatment: heart failure therapy		yes
38	Sata et al. ²⁹	2007	M	64	acute	RCIA	RCIV	open	Ligation of fistula, aorto-iliac bifurcation graft	uneventful, resolution of symptoms	yes

(Continued)

Table I. Continued

Case number	Author	Publication year	Sex	Age	Duration of symptoms	Artery	Vein	Technique	Treatment	Postop course	Survival
39	Pinto et al. ³⁰	2007		75		RCIA	RCIV	open	Aorto-iliac bifurcation graft	resolution of symptoms	yes
40	Goto et al. ³¹	2008	M	82	2 days	RCIA	RCIV	open	Direct closure fistula, aorto-iliac bifurcation graft. Reconstruction IMA	uneventful, resolution of symptoms	yes
41	Takano et al. ³²	2009	F	84	acute	RIIA	RCIV	open	Direct closure fistula, aorto-iliac bifurcation graft,	Uneventful, persistence of right foot edema	yes
42	Jakhere S. ³³	2010	M	75		LCIA	LCIV	none			no
43	O'Brien et al. ³⁴	2011	M	68		RCIA	RCIV	hybrid	See Table IV	uneventful, resolution of symptoms	yes
44	Patatas et al. ³⁵	2013	M	76		LIIA	LCIV	endovascular	See Table IV	uneventful, resolution of symptoms	yes
45	Rehman et al. ⁶	2014	M	58		RCIA	RCIV	endovascular	See Table IV	uneventful, resolution of symptoms	yes
46	Park et al. ³⁶	2015	M	64		LCIA	LCIV	endovascular	See Table IV	Type IIA endoleak, persistent fistula. Reintervention. See Table IV .	yes
47	Iijima et al. ³⁷	2015	M	86	acute	LCIA	LCIV	open	Direct closure fistula, aorto-iliac bifurcation graft, closure LIIA, reconstruction IMA	uneventful, resolution of symptoms	yes
48	Loos et al. ³⁸	2015	M	68	1 day	LCIA	LCIV	open	Direct closure fistula, aorto-iliac bifurcation graft	acute kidney failure, dialysis need, postanoxic encephalopathy	yes
49	Yamamoto et al. ³⁹	2015	M	52	acute	LIIA	LCIV	open		recovery without complications	yes
50	Hurndall et al. ⁴⁰	2017	F	73	6 hr	RCIA	RCIV	none	unfit for surgery		no
51	Caranzano et al. ⁴¹	2018	M	78	acute	LCIA	LCIV	none			no
52	Hachiro et al. ⁴²	2018	M	87	2 days	RIIA	RCIV	open	Ligation RCIA and RCIV, closure fistula. Femoro-femoral bypass.	uneventful, resolution of symptoms	yes
53	Borghese et al. ⁴³	2019	M	81	Acute	RCIA	RCIV	endovascular	See Table IV	uneventful, resolution of symptoms	yes
54	Doi et al. ⁴⁴	2019	M	41	acute	RCIA	RCIV	open	Aorto-femoral bifurcation graft		yes
55	Hanada et al. ⁴⁵	2019	M	44	several hours	RCIA	LCIV	open	Direct closure fistula, aorto-iliac bifurcation graft	uneventful, resolution of symptoms	yes

56	Rishi et al. ⁴⁶	2020	M	61		RCIA	RCIV	endovascular	See Table IV	Return of symptoms after 2 weeks: type IIIa endoleak, component separation. Reintervention. See Table IV	yes
57	Núñez et al. ⁴⁷	2020	M	74		RIIA	REIV	Endovascular start, conversion to open	Failed coiling, conversion to aneurysmectomy RIIA, direct closure of fistula, venous stent EIV	Uneventful, resolution of symptoms.	yes
58	Nishimura et al. ⁴⁸	2020	M	86	3 months	RCIA	RCIV	open	Fistula closure with patch, bifurcated graft	Almost resolution of symptoms, persistent occlusion CIV	yes
59	Sui et al. ⁴⁹	2021	M	64	1 day	RCIA	RCIV	endovascular	See Table IV	Type II endoleak: reintervention. See Table IV .	yes
60	Morton et al. ⁵⁰	2022	M	58		RCIA	RCIV	endovascular	See Table IV	Type II endoleak, reintervention. See Table IV	yes
61	Wang et al. ⁵¹	2022	M	61	2 days	RCIA	RCIV	endovascular	See Table IV	Type II endoleak, flow hemodynamically insignificant. Resolution of symptoms.	yes
62	Yoo, Y. ⁵²	2022	M	76	8 hr	RCIA	RCIV	endovascular	See Table IV	Type IIA endoleak, resolution of symptoms. No evidence of endoleak after 6 months.	yes
63	Gijssels et al.	2023	M	67	1 day	RCIA	RCIV	open	Direct closure fistula, aorto-femoral bifurcation graft	Progressive multisystem failure, hemodynamic instability.	No

Cases marked with * are included from Weimann et al.,³ cases marked with † are included from McAuley et al.¹

RCIA, right common iliac artery; LCIA, left common iliac artery; RIIA, right internal iliac artery; LIIA, left internal iliac artery; REIA, right external iliac artery; LEIA, left external iliac artery; RCIV, right common iliac vein; LCIV, left common iliac vein; RIIV, right internal iliac vein; LIIV, left internal iliac vein; REIV, right external iliac vein; RIA, right iliac artery; LIA, left iliac artery; RIV, right iliac artery; LIV, left iliac vein; IMA, inferior mesenteric artery.

Table II. Patient characteristics, treatment modalities, and outcomes

Characteristics	<i>N</i> = 63
Median age (years)	71 (63–76)
Sex	
Male	55
Female	4
Unspecified	4
Duration of symptoms	
≤ 24 hr	20
1 day–1 week	7
1 week–1 month	11
1 month–1 year	5
> 1 year	2
Unspecified	18
Diagnosis	
No preoperative diagnosis	5
CT scan	23
Angiography	19
Ultrasound	5
MRA	1
Unspecified radiography	1
Unspecified	9
Treatment	
Open	46
Endovascular	11
Endovascular + open	1
Converted	1
Unspecified	1
Deceased before surgery	3
Survival	
Yes	57
No	6

Data are presented as *n* or median (interquartile range).

CT, computed tomography; MRA, magnetic resonance angiography.

fistula and *'iliac aneurysm'* and excluded *'traumatic'* and *'iatrogenic'* in their title, abstract, or keywords. At last, the search terms for Web of Science included *'arteriovenous fistula'* and *'iliac aneurysm'* and excluded *'traumatic'*.

After exclusion of duplicates, a total of 461 search results were reviewed based on their title and abstract. Records that reported traumatic or iatrogenic cases, congenital aneurysms, mycotic aneurysms, and aneurysms secondary to tumor invasion, connective tissue disease, or arteritis were excluded. Furthermore, articles that were not in English, of which the full text was not available or abstracts from presentations were excluded. Finally, records were excluded if the majority of information was lacking, if they were not discussing iliac AVF or if there was involvement of the inferior vena cava or aorta. In total, 71 articles were reviewed for full

text using the same criteria and 43 records were withheld. Review of the references of those articles delivered 7 more articles. All were retrospective reports of cases or small literature reviews. Database searches were included until October 2022. The review was registered in the Prospero database under the Prospero ID CRD42022335318.

RESULTS

A total of 50 articles were included resulting in 62 cases of spontaneous iliac AVF, caused by rupture of an isolated iliac aneurysm into an iliac vein (Table I).^{1–3,6,7,9–52,53} The case described in this article was included, resulting in 63 cases.

Of the 63 patients, 55 were male and 4 female. In 4 cases, sex was not specified. The median age of patients was 71 years, ranging from 41 to 87 years (Table II). The triad described by McAuley et al. was only present in 32.7% of patients.¹ 47.3% of patients presented with only 2 of the 3 symptoms and 20.0% showed only 1. All patients had at least 1 symptom (Table III). The duration of symptoms until presentation was variable, ranging from less than an hour to 6 years.

In most patients, the iliac AVF was diagnosed preoperatively. In 23 cases, diagnosis was made with contrast-enhanced CT scan, 19 with angiography, 5 with ultrasound imaging, and 1 with magnetic resonance angiography. There were 6 cases in which no AVF was seen on the CT scan, but angiography did show a fistula. In 1 case, the type of imaging was unspecified. In 9 records, it was not specified how the diagnosis was made. In 5 cases, open surgery was initiated without being aware of the presence of an iliac AVF.

The level of the fistula was at the common iliac artery in 51 patients and at the IIA in 11 patients. In 1 case, the exact localization of the fistula was not specified.

Concerning treatment, the majority of patients underwent emergency surgery, with open surgery in 46 patients. In 13 patients, an endovascular intervention was initiated (Table IV). Seven of 13 endovascular interventions included endovascular abdominal aortic aneurysm repair, while in 2 patients a covered stent was placed in the iliac artery. In 6 of the endovascularly treated patients, embolization of the IIA at the side of the fistula was attempted, but was unsuccessful in 2 cases. One patient (case 43) was treated with an aorto-uni-iliac device in combination with an open femoro-femoral bypass.³⁴ Coiling of the fistula was attempted in 1 patient, but failed. The patient was considered unfit

Table III. Symptoms at presentation

Symptoms	1/3			2/3			3/3		
Sudden onset heart failure	+	—	—	+	+	—	+	78.2 (43)	
(unilateral) venous engorgement/leg ischemia	—	+	—	+	—	+	+	74.5 (41)	
Pulsatile abdominal mass	—	—	+	—	+	+	+	60.0 (33)	
	10.9 (6)	3.6 (2)	5.4 (3)	25.5 (14)	9.1 (5)	12.7 (<i>n</i> = 7)			
		20.0 (11)			47.3 (26)		32.7 (18)		

Data are presented as % (n). N = 55. 8 cases were excluded due to lack of information.

for surgery and solely received medical heart failure therapy (case 37).²⁸ In another patient (case 57), a conversion to open surgery was performed after a failed attempt to coil the IIA.⁴⁷

Five of the endovascularly treated patients had an uneventful postoperative course with complete resolution of their symptoms. One of them (case 35) was treated with a 2-staged approach where initially a covered stent was placed over the origin of the right IIA. Three weeks later, embolization of the aneurysm was performed using coils and thrombin and a venous bare metal stent was placed in the right common iliac vein to avoid embolization of the coils.²⁶ The second (case 43) received an aorto-uni-iliac stent graft with an occlusion device of the left common iliac artery and femoro-femoral bypass.³⁴ The third successful case (case 44) had a history of aorto-femoral bifurcated bypass and was treated with a covered venous stent in the left common iliac vein, coiling of the left internal iliac vein, and embolization of the native left external iliac artery.³⁵ The 2 remaining successful cases were treated with endovascular abdominal aortic aneurysm repair, one of which was combined with embolization of the right IIA (cases 45 and 53).^{6,43}

The most common reason for reintervention was endoleak with recurrence of symptoms. In 5 patients, a type II endoleak due to backflow from the IIA was seen, causing persistent AVF. Three needed a reintervention for coiling of the IIA that was successful in 2 of them. In the other 2 patients, no additional interventions were performed. All 5 patients had resolution of their symptoms eventually. In case 56, recurrent symptoms developed 2 weeks after treatment with an aortoiliac endograft and branched iliac graft, due to component separation of the bridging graft and the iliac branch graft. Revision surgery was necessary to place an additional iliac extension to bridge the main body and the iliac branch graft with an aortic cuff above the iliac branch flow divider. Additionally, venous stenting of the common iliac vein was performed using an

iliac extension limb. The patient recovered completely afterward.⁴⁶

In the open surgery group (46 patients), 3 patients died postoperatively (case 29, 30, and 63).^{22,23} Twenty patients had an uneventful postoperative course with resolution of their symptoms. In case 17, the patient suffered from a myocardial infarction perioperatively but recovered well.¹ Two patients had persistent leg edema. None of the patients required reintervention. Postoperative complications included thrombophlebitis of the leg (case 24), persistent occlusion of the common iliac vein (case 58), temporary impairment of liver function (case 21), and acute kidney failure (case 48).^{7,19,38,48} In 16 cases, no postoperative course was reported.

The overall mortality caused by the AVF was 9.5% (6 patients). Two patients died from a preoperative cardiorespiratory arrest and 1 patient was considered unfit for surgery so palliative treatment was started. The remaining 3 patients underwent open surgery, but died afterward due to progressive multisystem failure. Five of the deceased patients presented in shock or even cardio-respiratory arrest a few hours after start of their symptoms. The sixth had progressive symptoms of cardiac failure over a course of 3 years.

DISCUSSION

Acquired AVF of the iliac vessels can be divided into spontaneously arisen AVF and AVF secondary to vascular injury. The latter can be further divided into traumatic, most commonly stab (43%) or gunshot (32%) wounds,^{54,55} and iatrogenic (procedure-related),^{2,56} for example, after lumbar disk surgery or endovenous laser treatment of the great saphenous vein.^{56–60} In several cases, the fistula only becomes symptomatic several years after trauma.^{55,57,58,61–63}

For spontaneous iliac AVF, aneurysmal disease accounts for most of the cases. These aneurysms

Table IV. Overview of endovascular procedures and outcome

Case	Author	Artery	Vein	Treatment	Postop course
35	Char et al. ²⁶	RIIA	RCIV	1. Covered stent over origin RIIA 2. Access the aneurysm through the iliac vein to embolize the branches RIIA (unsuccessful) 3. (3 weeks later) Bare venous stent RCIV to prevent central embolization while placing coils and thrombin in the aneurysm	uneventful, resolution of symptoms
37	Tan et al. ²⁸	RIIA	RIIV	Failed attempt at coiling of the fistula	Heart failure therapy, outcome unknown
43	O'Brien et al. ³⁴	RCIA	RCIV	Aortic bifurcation diameter 19 mm, risk of compression seemed too high: 1. Aorto-uni-iliac device RIA occluding the RIIA 2. Occlusion device LCIA 3. Femoro-femoral bypass (8 mm polytetrafluoroethylene graft)	uneventful, resolution of symptoms
44	Patatas et al. ³⁵	LIIA	LCIV	1. Venous stenting across fistula opening 2. Embolization IIV 3. Embolization native LEIA (history: aorto-femoral bifurcation graft)	uneventful, resolution of symptoms
45	Rehman et al. ⁶	RCIA	RCIV	EVAR	uneventful, resolution of symptoms
46	Park et al. ³⁶	LCIA	LCIV	1. Embolization of LIIA 2. EVAR	Type IIA endoleak, persistent fistula. 1. Access through femoral vein 2. Coiling of a patent LIIA branch 3. Vascular plug at level of the fistula Resolution of symptoms
53	Borghese et al. ⁴³	RCIA	RCIV	1. Embolization RIIA 2. EVAR 3. iliac extension right side	uneventful, resolution of symptoms
56	Rishi et al. ⁴⁶	RCIA	RCIV	1. Right branched iliac graft 2. Embolization LIIA 3. EVAR 4. Bridging limb between main body and right iliac branch graft 5. Left iliac extension	Return of symptoms after 2 weeks: type IIIa endoleak, component separation. 1. Iliac extension graft main body to right iliac branch graft 2. Aortic cuff above iliac branch flow divider 3. Venous iliac extension limb REIV

(Continued)

Table IV. Continued

Case	Author	Artery	Vein	Treatment	Postop course
57	Núñez et al. ⁴⁷	RIIA	REIV	Failed attempt at coiling RIIA, conversion to open	
59	Sui et al. ⁴⁹	RCIA	RCIV	Impossible to enter aortoiliac grafts due to severe tortuosity Covered stent RIA	Type II endoleak with persistent fistula 1. Access through femoral vein. 2. Embolization RIIA with coils Resolution of symptoms. Type II endoleak
60	Morton et al. ⁵⁰	RCIA	RCIV	EVAR	1. Access through femoral vein 2. Impossible to cannulate RIIA Radiographic follow-up. Resolution of symptoms.
61	Wang et al. ⁵¹	RCIA	RCIV	1. Embolization RIIA 2. EVAR	Type II endoleak, flow hemodynamically insignificant. Resolution of symptoms.
62	Yoo, Y. ⁵²	RCIA	RCIV	1. failed attempt at embolization RIIA 2. EVAR 3. Right iliac extension 4. aortic extension cuff in the CIV over the venous fistula orifice	Type IIA endoleak, radiographic follow-up. Resolution of symptoms. No evidence of endoleak after 6 months.

RCIA, right common iliac artery; LCIA, left common iliac artery; RIIA, right internal iliac artery; LIIA, left internal iliac artery; LEIA, left external iliac artery; RCIV, right common iliac vein; CIV, common iliac vein; RIIV, right internal iliac vein; REIV, right external iliac vein; RIA, right iliac artery; EVAR, endovascular abdominal aortic aneurysm repair; LCIV, left common iliac vein.

are usually located deep in the pelvis, allowing silent growth and fistulization. They are often only diagnosed when secondary symptoms such as compression symptoms or cardiac failure appear.^{1,35,64}

Despite the classic triad that has been described in literature, only a third of patients present with this triad, as is shown in our review. The gravity of symptoms depends of the size and origin of the fistula.^{1,8} In up to 78.2% of patients with a spontaneous ilio-iliac AVF, the first complaints were related to heart failure. The diagnosis might be difficult, since this can be the only symptom.^{6,8,59} Patients may suffer from shortness of breath, chest pain, or palpitations. Lower limb ischemia might be present, caused by steal phenomenon or distal embolization of emboli arising inside the aneurysm,^{1,34,65} but venous engorgement and swelling of the leg from venous hypertension is more common and is seen in up to 75% of cases.^{2,8,65}

Acute onset of hemodynamic instability might occur in high output fistulas, such as an iliac aneurysm rupture into an adjacent vein.² Choosing the path of least resistance, the arterial blood flows

preferably into the venous system.⁶⁶ The increased venous return increases pulmonary blood flow, causing right heart decompensation.²⁷ The severity and progression of symptoms is related to the initial size and growth rate of the fistula, although a small fistula can cause heart failure as well.⁶⁷ In low output fistulas, right heart decompensation will develop progressively and symptoms such as ascites and hepatomegaly can occur. If the cardiac failure becomes manifest, patients can present with prerenal kidney failure, resulting in anuria or oliguria.^{6–8,25,27,66,68}

Other, more uncommon symptoms are hematuria or rectal bleeding, supposedly caused by pelvic venous congestion.^{2,24} Another unusual complication is pulmonary embolism (paradoxical embolism).^{16,24} Furthermore, hydroureteronephrosis due to compression of the ureter by the enlarged iliac vessels has also been reported.³ In some patients, the rapid flow in the fistula can cause anemia, as was the case in our patient.^{69,70}

CT angiography is most commonly used for the diagnosis of iliac AVF, but is sometimes insufficient.

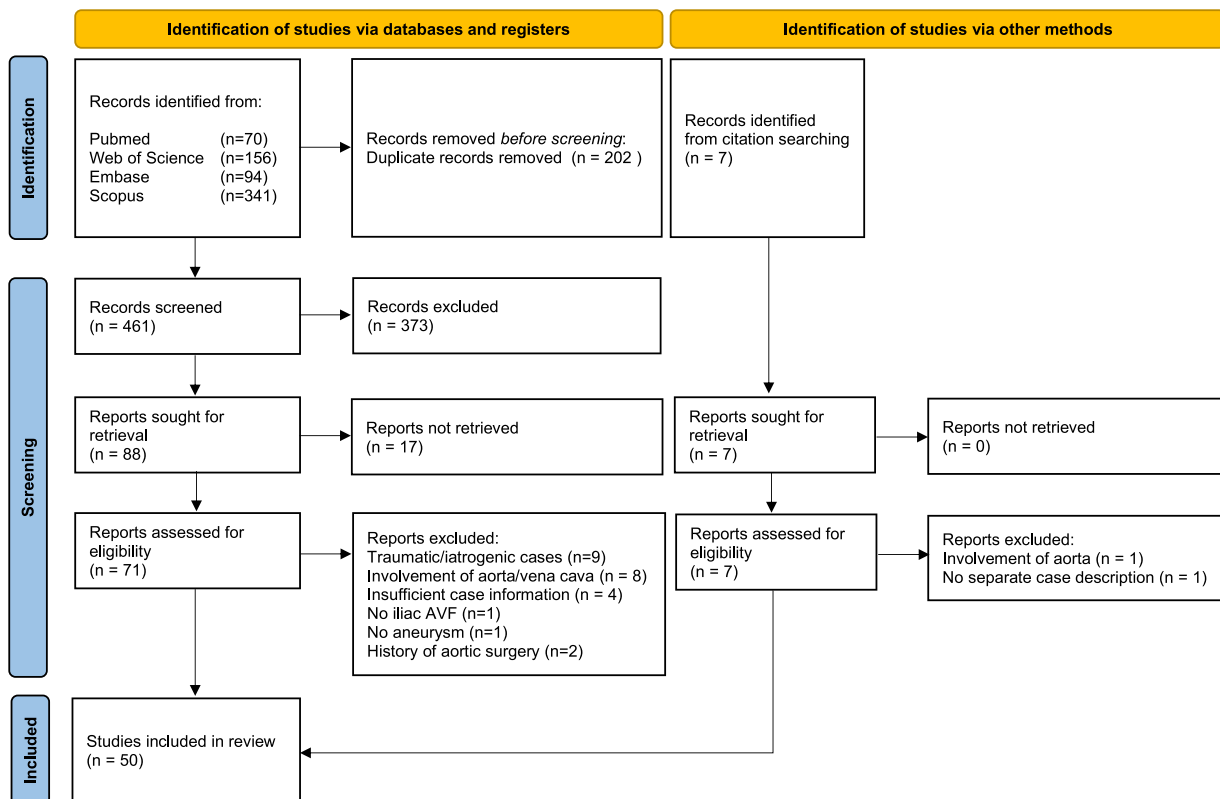


Fig. 3. Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart. A total of 461 records were screened.

Angiography is more accurate for smaller fistulas and might show an AVF that could not be diagnosed using CT scan, as in 6 of the included cases.^{1,8,37} Characteristic signs of AVF are saccular aneurysmal dilatation of the draining vein and rapid enhancement of the iliac vein and vena cava during the arterial phase.^{64,71} Determining the exact location of the fistula is possible due to reconstruction techniques such as volume rendering or virtual angioscopy.³¹

As heart failure symptoms often predominate, many patients get a complete cardiac work-up. Clinical examination can reveal a cardiac murmur. Chest X-ray and transthoracic echocardiography might demonstrate signs of pulmonary hypertension and dilation of the ventricles, suggesting biventricular overload. Right heart catheterization revealing elevated venous pressure and a low arterial-mixed venous oxygen differential can raise suspicion of an extracardiac AV shunt.^{6,7,25,59,60}

The treatment of iliac AVF is mainly surgical, with closure of the fistula while maintaining arterial and venous perfusion. Most experience is based on open surgery. To avoid embolization, careful dissection and early clamping of both feeding artery and

vein is recommended.^{8,67} Because of enlargement of surrounding veins, dissection might be complicated by extensive bleeding.⁶³ Once the feeding artery and draining vein are clamped, the aneurysm sac is opened with closure of the fistula and vascular reconstruction. Usually, a bifurcated aortic graft is required for iliac AVF originating in an aneurysm.^{1,2} Ureteral damage is a known risk when performing surgery on the iliac vessels, but no ureteral damage has been reported in the cases that were included in this review.

Modern-day endovascular techniques are also used to close iliac AVF. Major benefits of this approach are the limited bleeding risk and less invasive approach in this patient population with high cardiovascular risk. Furthermore, in experienced hands, the intervention takes less time and the fistula can be excluded faster when compared to open surgery. However, it might be more difficult to maneuver through the aneurysm and the technique is not always successful, as is shown in this review. In case of severe tortuosity or calcification of the vessels, an endovascular approach might not be possible. The exact endovascular treatment

depends on the exact localization of the fistula and the presence of landing zones. Literature reports exclusion of the orifice of the fistula with a covered arterial stent in case of suitable proximal and distal landing zones. Otherwise, a bifurcated stent graft should be considered.^{34,71} Long-term results are however lacking and the risk of a potential endoleak is not negligible.^{4,37,59,60} In addition, venous stenting might be considered to prevent central embolization or thrombosis caused by compression.^{35,54,58,63,64} The indications for venous stenting that were identified in this systematic review included persistent fistula opening and prevention of central embolization after coiling and injecting thrombin into the aneurysmal sac. It should however be taken into account that application of a venous stent can hamper access to the aneurysmal sac in case of reintervention for endoleak.

In this literature review, only 5 of 11 endovascularly treated patients had an uneventful success. Six of 11 patients showed an endoleak at postoperative control and 4 of them needed an additional intervention. In case of bilateral iliac aneurysms, at least 1 hypogastric artery should be preserved; hence, an open approach could be more suitable.⁷² It is important to be aware of the anatomy of the aneurysm and fistula opening to adapt the surgical approach.

During the perioperative and postoperative course, an abrupt rise in blood pressure can be observed, caused by the prompt closure of the AVF.⁶⁶ This resolves after a few days, when the volume overload diminishes. Excessive diuresis of 12L over 2 days has been reported.⁸

Outcomes of surgical interventions are good. The reported mortality rates are low, provided there was an accurate preoperative diagnosis and immediate surgical measures were taken.^{1,8,27,67} In this review, only 3 of 60 patients who underwent surgery did not survive. Postoperatively, most patients experience a complete resolution of their symptoms and no recurrence of the AVF.^{1,7,24,67}

CONCLUSION

Iliac AVF should be suspected in case of symptoms of high output cardiac failure combined with an abdominal pulsatile mass or unilateral leg engorgement. Patients may present with various symptoms, making the diagnosis difficult. CT angiography is the gold standard for diagnosis, showing rapid enhancement of the inferior vena cava. Additional angiography might be necessary to diagnose smaller AVF. Elective repair of these fistulas shows excellent

results, but understanding the anatomy of the AVF is key in tailoring the surgical strategy. Endovascular repair can be performed, but the short-term and long-term results are still suboptimal compared to open procedures.

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