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# CONGESTIVE HEART FAILURE AND LOWER EXTREMITY EDEMA AS LATE SEQUELAE OF UNRECOGNIZED ILIAC ARTERIOVENOUS **FISTULA**

Kongestivno zatajenje srca i edem donjeg ekstremiteta kao kasni nastavak neprepoznate ilijačne arteriovenske fistule

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#### **Abstract**

We report a case of a female patient with a large volume iliac arteriovenous fistula. She presented with severe congestive heart failure and unilateral leg edema. Six years before present admission she underwent surgery because of the lumbar disc protrusion. Diagnosis was delayed due to the overshadowing symptoms of coexistent aortic valve insufficiency. Contrast-enhanced multislice computed tomography with threedimensional reconstruction confirmed the diagnosis. The fistula was closed by using an open surgical approach, and the patient completely recovered. Severe late consequences of unrecognized arteriovenous fistula are emphasized. Causes for delayed recognition of underlying pathology, as well as diagnostic and treatment options are discussed.

## **Key words**

aortocaval and iliac, arteriovenous fistula, congestive heart failure, leg edema, spinal surgery

### Sažetak

Prikazujemo slučaj bolesnice s visokoprotočnom ilijačnom arterijsko-venskom fistulom. Bolesnica se prezentirala teškim kongestivnim zatajenjem srca i unilateralnim edemom noge. Šest godina prije sadašnjeg prijema, bolesnica je operirana zbog protruzije lumbalnog intervertebralnog Dijagnoza arterijsko-venske fistule je postavljena kasno, zbog prikrivenosti tegoba pridruženom insuficijencijom aortalnog zaliska, a potvrđena je spiralnom kompjutoriziranom tomografijom kontrastom i trodimenzionalnom rekonstrukcijom. Fistula je zatvorena otvorenim kirurškim pristupom. Bolesnica se potpuno oporavila nakon operacije. U ovom prikazu ističemo teške kasne posljedice neprepoznate arterijsko-venske fistule i razmatramo uzroke kasnog postavljanja dijagnoze, kao i različite mogućnosti dijagnostike i liječenja.

## Ključne riječi

aortokavalna i ilijačna, arterijsko-venska fistula, srčana dekompenzacija, edem noge, spinalna kirurgija

#### Introduction

Iliac arteriovenous fistulas (AVF) are rare and even less frequently reported than aortocaval fistulas [1, 2]. The most common etiological factors include rupture of the iliac artery aneurysm into the adjacent vein, followed by penetrating trauma and iatrogenic causes like lumbar surgery [1–4]. Typical clinical features include lumbar pain, high cardiac output, unilateral lower extremity edema, leg ischemia or intermittent claudication and a machinery-like bruit. A palpable thrill over the fistula site is usually present [1, 5]. If diagnosis is delayed, severe late complications like heart failure, renal and hepatic insufficiency may occur [1, 6–9]. Therefore, early diagnosis and treatment are mandatory. Different noninvasive as well as invasive diagnostic procedures are used, but precise delineation of the fistula site is the goal of utmost importance. Treatment modalities depend on the patient's and local AVF characteristics. Successful endovascular as well as open repairs have been reported.

## **Case report**

A 57-year-old woman was referred due to the progression of exertional dyspnea, left leg edema and lumbar pain. Six years before present admission she underwent surgery because of disc protrusion at the

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L4-L5 level. Two years after lumbar surgery she

underwent coronarography because of chest pain and suspected coronary artery disease (CAD). While the diagnosis of CAD was not confirmed, the diagnosis of the mild aortic valve stenosis (AVS) was established and the patient has been followed by a cardiologist ever since. Physical examination at present admission revealed distended superficial neck and abdominal veins, a massive thrill on abdominal palpation and a continuous machinery-like bruit over the lower abdominal wall. Apart from the left leg edema, the calf skin induration and hyperpigmentation were also present (Fig. 1). Chest X-ray showed cardiomegaly and increased pulmonary vascular markings (Fig. 2). Echocardiography showed hyperdynamic motion of the left ventricular (LV) wall and aortic valve stenosis, with a systolic gradient of 58 mmHg. Elevated total bilirubin (43 µmol/L) and gamma-glutamyltransferase (96 U/L) indicated liver stasis. Multislice computed angiography (MSCTA) with a three-dimensional (3D) reconstruction demonstrated a high-flow AVF of 11 mm in diameter between the right common iliac artery and the left common iliac vein. The inferior vena cava (IVC) was dilated to 55 mm and both common iliac veins were also dilated. The right common iliac artery was enlarged with the maximum transverse diameter of 18.8 mm (Fig. 3). Hepatic veins and pulmonary arteries were also dilated. Due to the progression of severe symptoms, we decided to intervene immediately. Because of the hemodynamic characteristics of the AVF, local anatomy and patient's condition, we decided to perform an open surgery. Full invasive monitoring was established and a cell-saver device was obtained. Preoperative hemodynamic parameters were: cardiac output (CO) 13.2 L/min; cardiac index (CI) 7.5 L/min/m2; stroke volume (SV) 212.9 ml/beat, mean pulmonary artery pressure (PAP) 47 mmHg; and pulmonary artery occlusion pressure (PAOP) 17 mmHg. Midlaparotomy was performed. The AVF was located between the proximal portion of the right common iliac artery and the left common iliac vein. The arterial and venous walls were tightly approximated with dense fibrotic tissue in between, so we decided not to separate the vascular structures. Thorough exploration of the abdominal cavity was performed as to reveal any additional pathology. Heparin was administered and we clamped the aorta and the distal common iliac arteries away from the neighboring dilated veins. The inferior vena cava and the common iliac veins were also clamped. The common iliac artery was opened longitudinally, the fistula identified and closed with a continuous 4-0 prolene suture. The suture line was reinforced with Teflon pledgets. The arterial wall was extremely friable and we decided to perform an arterial reconstruction with "Y" Dacron silver graft (16x8 mm). The proximal anastomosis was created at the level of the infrarenal aorta, while the distal anastomoses were performed at the level of the distal common iliac arteries. The immediate postoperative values of the hemodynamic parameters improved: CO was 5 L/min, CI 2.9 L/min, SV 94.3 ml/beat, mean PAP 20 mmHg and PAOP 35 mmHg. The leg edema rapidly decreased within several days and the patient was released from the hospital on the 12th postoperative day. During the eight-month follow-up period, her cardiac and liver function significantly improved and she regained complete walking ability.

#### Discussion

Clinical manifestations of an aortocaval and iliac AVF may comprise a wide array of symptoms. In the majority of reported cases, aortocaval fistulas resulted from an aortic aneurysm rupture. Such patients have usually had an acute presentation due to the large volume of shunting blood followed by cardiac overload. Most patients with a large volume arteriovenous fistula manifest progressive dyspnea on exertion, an abdominal bruit and lower back pain. Because of the large shunting volume of the AVF, high-output cardiac failure may occur. Dilatation of the inferior vena cava and iliac veins as well as lower extremity edema are often associated [1, 5, 6, 10]. Liver and renal failure have been reported as a consequence of the acutely emerging aortocaval AVF [1, 3,6-9]. Apart from an acute presentation of the large volume AVF, in some cases the symptoms may arise gradually over time, due to the progressive fistula enlargement. Such clinical situations may emerge after vascular lesions during lumbar disc surgery, if the initial bleeding is overlooked. In those patients, the diagnosis of AVF may be delayed. In a review written by Brewster et al., the interval from the presumed occurrence of vascular lesion to the established diagnosis of AVF ranged from several hours to eight years. The authors reported that the most prominent symptoms of AVF were back pain (70%) and abdominal bruit (80%). Severe leg edema was the primary manifestation in eight out of 20 cases, while congestive heart failure occurred in only seven (35%) patients. Four AVFs resulted from a previous lumbar surgery [1]. The first AVF as the sequel of lumbar surgery was reported by Linton and White in 1945 [11] and more cases were reported afterwards. Vascular lesions during lumbar surgery usually occur during instrumentation of the disc space at the levels L4-L5 and L5-S1 [4, 11-13]. The vessels at this level are particularly vulnerable to injury because of their relative immobility and proximity to the spine when the patient is in prone position. In our patient, the AVF occurred at the level of L4-L5 vertebrae, corresponding precisely with the site of the previous lumbar surgery. The location of the AVF and the patient's history led us to presume that the initial vascular lesion had resulted from a lumbar disc surgery six years before present admission. Sometimes, the existing symptoms of AVF

may be misinterpreted or overshadowed by symptoms of an associated pathology. In the report of a patient with multiple organ failure secondary to iliac aneurysm rupture into the adjacent vein, the diagnosis was delayed as the multi-organ derangement was attributed to sepsis [6]. Another case report was related to a 71-year-old woman treated for unilateral leg edema. Because of the symptoms progression, an additional diagnostic evaluation had been performed that revealed an aortocaval fistula. The diagnosis was missed in the early phase of the disease primarily because of the low shunting volume through the AVF [5]. In our patient, the aortic valve stenosis certainly overshadowed the existing symptoms of the AVF. In order to avoid severe late complications, early diagnosis and treatment of AVF are essential. Over years, conventional angiography was most commonly used to define the diagnosis of AVF. Introducing the spiral and multislice computed tomography, the evaluation of a suspected aortocaval and iliac AVF is possible using **MSCT** angiography Gadolinium-enhanced [3]. magnetic resonance angiography has recently been introduced as a precise diagnostic method while avoiding iodinated contrast agents [13]. Using contrast enhanced MSCT angiography in our patient, early synchronous and equivalent enhancement of the iliac veins, inferior caval vein and aorta was demonstrated, thus indicating the presence of the AVF. The precise location of the fistula, as well as clear visualization of all surrounding structures, was demonstrated after 3D reconstruction. Although rare, the majority of literature data have reported on open surgery for patients with AVF that manifested the signs of acute or progressive deterioration [1, 3, 9, 10]. Open surgery in such patients is often associated with massive bleeding and with significant hemodynamic disturbances. However, if anatomic features and the patients condition are favorable, endovascular treatment may be the first line therapeutic option [2, 12, 13]. In our patient, the diameter of the AVF was large and the degree of shunting was enormous. The inflow iliac artery was enlarged to the aortic bifurcation. The patient also exhibited severe progressive deterioration. Taking all above into consideration, we decided to perform an open surgery without any delay. With an open approach we achieved complete intraoperative control of all involved vascular structures. Clamping the adjacent arteries and veins proximal and distal to the fistula site provided a bloodless operative field and a precise fistula closure, without the need to hurry. Due to chronic inflammatory and adhesive changes of the tissue surrounding the AVF, vascular anastomoses were created within the segments of the normal arterial wall.

## Conclusions

An unrecognized AVF may lead to severe life threatening complications. In the presence of an acute large volume AVF, followed by typical symptoms, the diagnosis is usually established early. On some occasions, like lumbar surgery, early symptoms after a vascular lesion may be discrete and such lesions are easily overlooked. In patients who develop cardiac symptoms after lumbar disc surgery, diagnostic work-up for revealing the AVF is mandatory. Special attention has to be given to patients with preexisting cardiac pathology, thus masking the symptoms emerging from the AVF. Anatomic and pathophysiologic characteristics of the fistula as well as the patient's general condition are essential with regard to treatment modality. A less invasive endovascular approach is recommended as the first-line treatment option in appropriate cases. An open surgery should be considered in some unstable patients with complex pathology and in those with an unfavorable anatomy not amenable for endovascular access.



Figure 1. Cardiomegaly and increased pulmonary vascular markings on the preoperative chest X-ray.



Figure 2. Edema, varicosities and skin discoloration of the patient's left calf.

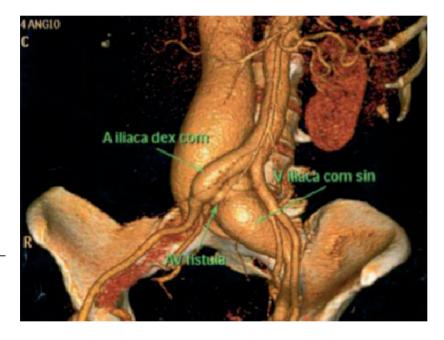


Figure 3. MSCTA with 3D reconstruction, demonstrating arteriovenous fistula between the right common iliac artery ("A iliaca dex com") and left common iliac vein ("V iliaca com sin").

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