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Unusual Case of Pulmonary Hypertension due to Acquired Arteriovenous Fistula

Edinilmiş Arteryovenöz Fistüle Bağlı Nadir Bir Pulmoner Hipertansiyon Vakası

ABSTRACT

Pulmonary hypertension is a disease process affecting pulmonary circulation and is defined by an increase in pulmonary artery pressure subsequently causing right ventricular failure. Vascular complications, including arteriovenous (AV) fistula, are recognized, but are uncommon complications of spinal surgery. AV fistula increases venous return to the right heart and can promote a volume overload related high-output cardiac state, pulmonary hypertension, and right heart failure. Hereby, we describe a rare pulmonary hypertension case with severe right heart failure, lower leg edema, and progressive dyspnea caused by an AV fistula between the left common iliac artery and vein as a complication of a lumbar spinal/disk surgery. Pulmonary hypertension was confirmed by hemodynamic assessments and the etiology was established by both abdominal computed tomography and conventional peripheric angiography. After closure of the AV-fistula by stent-graft implantation, the right heart failure resolved completely.

Keywords: Arteriovenous fistula, endovascular, heart failure, lumbar surgery, pulmonary arterial hypertension, tricuspid regurgitation

ÖZET

Pulmoner hipertansiyon, pulmoner dolaşımı etkileyen, sağ ventrikül yetmezliğine neden olan pulmoner arter basıncındaki artışla tanımlanan bir hastalık süreci olarak tanımlanır. Arteriovenöz fistül dahil vasküler komplikasyonlar spinal cerrahinin nadir görülen komplikasyonlarıdır. Arteriovenöz fistül sağ kalbe venöz dönüşü artırıp hacim yüklenmesiyle ilişkili yüksek debili kardiyak duruma neden olarak, pulmoner hipertansiyona ve sağ kalp yetmezliğine yol açabilir. Bu makalede lomber spinal/disk cerrahisinin bir komplikasyonu olarak sol ana iliak arter ile ven arasındaki arteriyovenöz fistülün neden olduğu ciddi sağ kalp yetmezliği, alt bacak ödemi ve progresif dispne ile seyreden nadir bir pulmoner hipertansiyon olgusu sunulmaktadır. Pulmoner hipertansiyon hemodinamik değerlendirmelerle doğrulanmış ve etiyoloji hem abdominal bilgisayarlı tomografi hem de konvansiyonel periferik anjiyografi ile belirlenmiştir. Olguda AV-fistülün stent-greft implantasyonu ile kapatılmasından sonra sağ kalp yetmezliği tamamen düzelmiştir.

Anahtar Kelimeler: Arteriovenöz fistül, endovasküler, kalp yetersizliği, lumbar cerrahi, pulmoner arteriyel hipertansiyon, triküspid yetmezliği

CASE REPORT
OLGU SUNUMU

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Diagnosis of pulmonary hypertension (PH) is generally delayed due to non-specific symptoms in initial presentation of patients. Detection of PH in early stages may reveal possible reversible etiologies and early introduction of specific therapies can prevent or slow the disease progression. However, in overlooked and untreated cases of PH; clinical progression inevitably results impaired exercise tolerance of the patient and right heart failure or in worst case scenario sudden cardiac death may occur.[1]

A detailed patient history, comprehensive physical examination, and guideline-based algorithmic diagnostic testing are essential for the diagnosis and management of PH. In the current diagnostic evaluation concept; identification and elimination of common PH etiologies is a must, to reach idiopathic pulmonary arterial hypertension diagnosis. Another focus of this approach is to define potentially reversible etiologies and initiate appropriate treatments before permanent changes occur in pulmonary vascular bed. In fact, reversible causes of PH are uncommon and include chronic thromboembolic disease which can be cured by pulmonary endarterectomy or correctable several left heart diseases such as mitral stenosis, cor triatriatum, or obstructive left atrial myxoma and

some congenital heart defects. Acquired systemic arteriovenous (AV) fistulas are also a less recognized but reversible etiology of secondary PH. Timely identification and treatment of this rare cause of PH can resolve patients' symptoms and may improve their quality of life.[2-4]

Hereby, we describe a rare PH case with severe right heart failure, lower leg edema, and progressive dyspnea caused by an AV fistula between the left common iliac artery and vein as a complication of a lumbar spinal/disk surgery. PH was confirmed by hemodynamic assessments and the etiology was established by both abdominal computed tomography and conventional peripheric angiography. After closure of the AV-fistula by stent-graft implantation, the right heart failure resolved completely.

Case Report

A 60-years-old female patient was referred for suspicion of chronic thromboembolic pulmonary hypertension (CTEPH). She had progressive exertional dyspnea and peripheral edema. Functional class was New York Heart Association (NHYA) Class III and initiation of symptoms coincided with lumber disk surgery performed 9 months ago without eventful recovery and without prolonged immobilization. Medical history was unremarkable except systemic hypertension and she denied smoking and any recreational drug use. Physical examination revealed uncontrolled blood pressure (180/70 mmHg) and 2/6 grade systolic murmur in tricuspid and mitral areas and bilateral Grade 3+ pitting edema. She did not have cyanosis or clubbing. Electrocardiography showed normal sinus rhythm, left axis, and 1° AV block. Echocardiographic examination revealed slightly hyperkinetic left ventricular function, mildly dilated left atria, and mild mitral regurgitation. Right heart chambers were mildly dilated with apparent septal bounce. Estimated pulmonary arterial pressure was calculated as 86 mmHg over severe tricuspid regurgitation.

Chest X-ray showed cardiomegaly with clear lung areas (Figure 1). Laboratory investigations including PH-related tests (Hepatitis and HIV serology and ANA tests) were unremarkable. However, BNP value was high (734 pg/mL). Pulmonary function tests indicated mild restrictive pattern and lung diffusion capacity for carbon monoxide was normal. Lung perfusion/ventilation spect showed mismatch subsegmental defect at left apicoposterior segment while no embolic defect was detected by pulmonary computed tomography (CT) angiography revealing biatrial dilatation and cardiomegaly. Bilateral geographic paranchymal pattern was observed and the main pulmonary trunk was normal (25 mm).

According to PH work-up requirements, cardiac catheterization was performed revealing precapillary PH (mean PAB: 25 mmHg; PCWP:9 mmHg, pulmonary vascular resistance (PVR): 1.64

ABBREVIATIONS

AV	Acquired systemic arteriovenous
CT	Computed tomography
CTEPH	Chronic thromboembolic pulmonary hypertension
NHYA	New York Heart Association
PH	Pulmonary hypertension
PVR	Post-void residual
PVR	Pulmonary vascular resistance

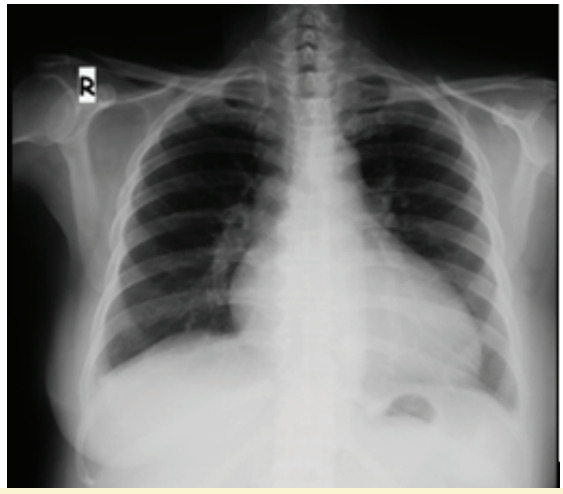

Figure 1. Pre-operative chest radiograph.

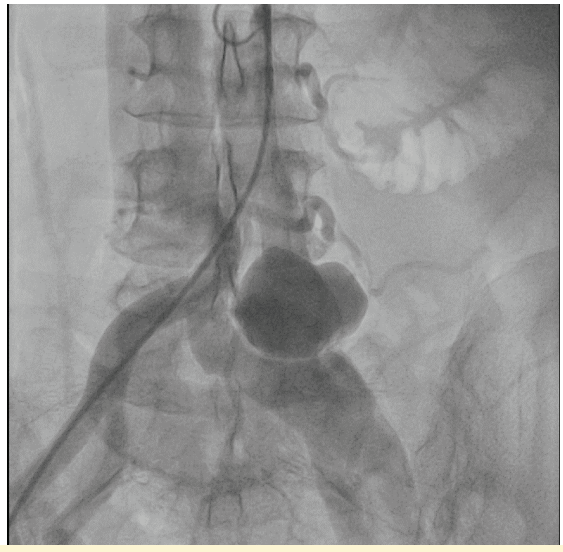

Figure 2. Peripheral angiography. Arteriovenous fistula between left common iliac artery and vein.

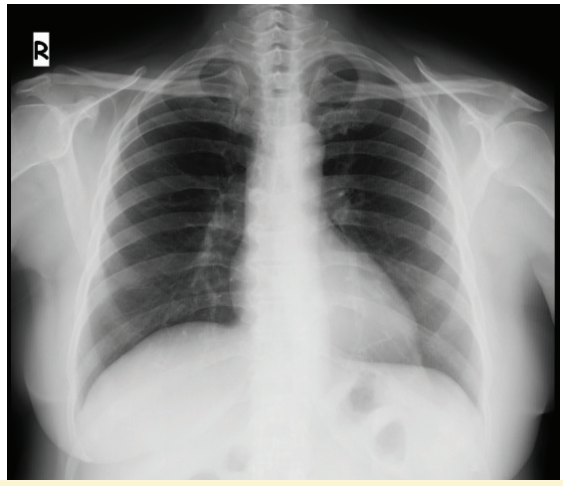

Figure 3. Post-operative chest radiograph.

Table 1. Catheterization Results

Oxymetric run		Pressures (mmHg)		Calculations	
Right Femoral vein	82.6	RA	(3)	Qp	9.76 L/min
Left common iliac vein	96.8	RV	50/0/3	Qs	6.68 L/min
VCI-inferior (subdiaphragmatic L4-L5 level)	95.2	PA	45/15 (25)	Qp/Qs	1.46
VCI-superior (diaphragmatic level)	93.5	PAWP	(9)	PVR	1.64 wu
VCS	78.7	LV	200/0/12	SVR	18.41 wu
MvO_2 (*)	82.6	Aorta	197/72 (126)	CI	3.86 L/min/m^2
RA mid-level	85.7				
RV apex	86.5			BSA	1.73 m^2
Main PA	87			BMI	32 kg/m^2
Aortic root	99.5				
SpO_2	99.0				

(*) As oximetric step-up was detected between right femoral vein and subdiaphragmatic inferior vena cava; Flamm formula was used to define mixed venous oxygen (MvO_2) value, utilizing SVC and right femoral vein oxygen saturations. VCI, Vena cava inferior; VCS, Vena cava superior; MvO_2, Mixed venous oxygen saturation; RA, Right artia; RV, Right ventricle; PA, Pulmonary artery; PAWP, Pulmonary arterial wedge pressure; LV, Left ventricle; Qp, Pulmonary output; Qs, Systemic output; Qp/Qs, Shunting ratio; CI, Systemic cardiac index; PVR, Pulmonary vascular resistance; SVR, Systemic vascular resistance, BSA, Body surface area; BMI, Body mass index.

Woods unit, Pulmonary output (Qp):9.76 L/min, Systemic output (Qs):6.68 L/min, Shunting ratio (Qp/Qs):1.46, and normal cardiac index (3.86 L/min/m^2) (Table 1). Coronary angiography was normal while peripheral angiography indicated significant shunting through AV fistula between the left common iliac artery and vein (Figure 2).

Eventually precapillary PH was attributed to the atriovenous fistula and the fistula was closed percutaneously in another session. The patient's symptoms disappeared soon after the closure with full recovery of clinical findings and echocardiographic improvement. Cardiothoracic ratio was normalized and echocardiography showed mild tricuspid regurgitation with an estimated PASB of 36 mmHg at 7 months. At long-term follow-up, the patient had no complaints and her functional was still NYHA Class I with normal pulmonary artery pressures estimated by echocardiography (Figure 3).

Discussion

This report describes a rare PH case with severe right heart failure, lower leg edema, and progressive dyspnea caused by an AV fistula between the left common iliac artery and vein as a complication of a lumbar spinal/disk surgery. Pulmonary hypertension was confirmed by hemodynamic assessments and the etiology was established by both abdominal computed tomography and conventional peripheric angiography. After closure of the AV-fistula by stent-graft implantation, the right heart failure resolved completely similar to previously reported cases.[5,6]

Although the patient was referred for CTEPH investigation with a documented mismatch defect on lung V/Q scan; according to our initial evaluation, the presence of PH and etiological evidence was clear for us: Because only one minor subsegmental mismatch defect (in the context without any other clues indicating venous thrombosis) was far away from explaining the whole clinical picture as CTEPH, while the symptoms occurring just after the spinal surgery and rapidly progressing to right heart failure in 9 months and significant trill and bruit in abdominal examination suggested us the possibility of an AV-fistula. Therefore, besides a work-up for CTEPH evaluation, we also performed CT imaging confirming AV fistula before catheterization.

The occurrence of an A-V shunt with PH after spinal disk surgery has rarely been reported in the literature, and in all of them, high-output cardiac failure was present.[5,6] In our case, abnormal pulmonary vascular resistance (PVR) value which does not exceed 2-wood unit limit indicates that pulmonary vascular disease has not been established yet; and also high-output cardiac failure was absent as cardiac index was almost at upper limit of normal. However, this does not warrant exclusion of the diagnosis and can be due to utilization of different measurement methods (Fick method for the given case) and for our case, effective pre-treatment with loop diuretics before catheterization may have partially improved hemodynamics by alleviating volume overload. A systemic A-V shunt after surgery can rarely cause PH, and this has to be considered as a rare cause of secondary PH, which easily may be overlooked in conventional differential diagnoses of PH.

Although treatment for most acquired A-V shunts is surgical, recently endovascular techniques have been recommended, lowering the high morbidity and mortality related to conventional repair. When PH of an uncertain etiology occurs in patients previously submitted to lumbal surgery, a careful clinical examination (including abdominal oscultation) can lead to a correct diagnosis of an iatrogenic A-V fistula, and surgical or endovascular correction usually results in the restoration of the normal cardiac functions.[5]

Conclusion

Acquired AV fistulas are rare causes of high-output cardiac failure and may present as PH of uncertain cause. Close attention to a history of invasive procedures or penetrating injuries and a careful

physical examination is of paramount importance in recognizing this entity.[6]

Informed Consent: Written informed consent was obtained from the patient for their anonymized information to be published in this case report.

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Author Contributions: Design – M.O.; Writing – İ.B.

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