

Agitated saline contrast use in a case with peripheral pulmonary artery stenosis

Periferik pulmoner arter darlığı olan bir olguda kontrast ajite salin kullanımı

Zafer Işılak, M.D., Mehmet Uzun, M.D., Fethi Kılıçaslan, M.D., Ömer Uz, M.D.

Department of Cardiology, Haydarpaşa Training Hospital, Gülhane Military Medical School, İstanbul

Summary – We present a case of peripheral pulmonary artery stenosis that was incidentally detected by agitated saline contrast study. A 19-year-old male patient underwent cardiologic examination to determine suitability for military service. He had exertional intolerance since early childhood. Physical examination showed a murmur over the entire right hemithorax. Echocardiography showed moderately enlarged right ventricle, right atrium, and main pulmonary artery, and color Doppler showed mild tricuspid regurgitation. Right ventricular systolic pressure was estimated as 55-60 mmHg from the tricuspid regurgitation jet. For further evaluation of the systolic murmur, agitated saline contrast echocardiography was performed. During continuous wave Doppler examination while there were remnants of bubbles in the right heart and pulmonary vascular bed, a systolodiastolic flow with a peak gradient of 30 mmHg was noted. After disappearance of the bubbles, the signal was not detectable. Repeat agitated saline contrast examination again showed a gradient of 35 mmHg. A stenosis in the distal branches of the right pulmonary artery was suspected. Finally, computed tomography revealed multiple stenoses in the pulmonary vascular bed. To our best knowledge, this is the first case in which agitated saline contrast examination enabled the diagnosis of peripheral pulmonary artery stenosis.

Agitated saline contrast echocardiography is a safe, convenient and easy method in evaluating many cardiac entities. Due to elimination of saline contrast in pulmonary vascular bed, its use is generally limited to abnormalities of the right side of the heart.^[1] Among these conditions, persistent vena cava superior and patent foramen ovale are the leading ones. It is also used to estimate pulmonary artery

Özet – Bu yazıda, ajite salin kontrast ekokardiyografi sırasında raslantısal olarak saptanan periferik pulmoner arter darlığı sunuldu. On dokuz yaşında erkek hasta, askerlik hizmetine uygun olup olmadığını belirlemek için kardiyolojik açıdan değerlendirildi. Hastada çocukluğundan beri egzersiz intoleransı vardı. Fizik muayenede tüm sağ hemitoraks üzerinde bir üfürüm duyuldu. Ekokardiyografik incelemede, sağ ventrikül, sağ atriyum ve ana pulmoner arter orta derecede genişlemiş izlendi. Renkli Doppler görüntülemeye hafif triküspit yetersizliği vardı. Triküspit yetersizlik akımından ölçülen sağ ventrikül sistolik basıncı 55-60 mmHg idi. Sistolik üfürümün aydınlatılması için hastaya ajite salin kontrast ekokardiyografi yapıldı. Sürekli dalga Doppler incelemesi sırasında, sağ kalpte ve pulmoner vasküler yatakta henüz daha kabarcıklar varken, zirve farkı 30 mmHg olan sistolik ve diyastolik bir akım görüldü. Kabarcıklar kaybolduğunda sinyal de kayboldu. Tekrarlanan ajite salin kontrast enjeksiyonunda yeniden 35 mmHg'lik bir fark ortaya çıktı. Bu bulguyla, sağ pulmoner arterin distal dallarında darlıktan şüphelenildi. Bunun üzerine yapılan bilgisayarlı tomografide pulmoner vasküler yatakta darlıklar görüldü. Bildiğimiz kadarıyla, olgumuz, ajite salin kontrast incelemesi sayesinde periferik pulmoner arter darlığı tanısı konan ilk olgudur.

pressure in cases with insufficient Doppler signal of tricuspid regurgitation.^[1] In this paper, we present a case of peripheral pulmonary artery stenosis that is seldom encountered in routine clinical practice and difficult to diagnose with routine echocardiographic examination. The use of agitated saline contrast echocardiography made an important contribution to the diagnosis.

Received: March 16, 2011 Accepted: July 20, 2011

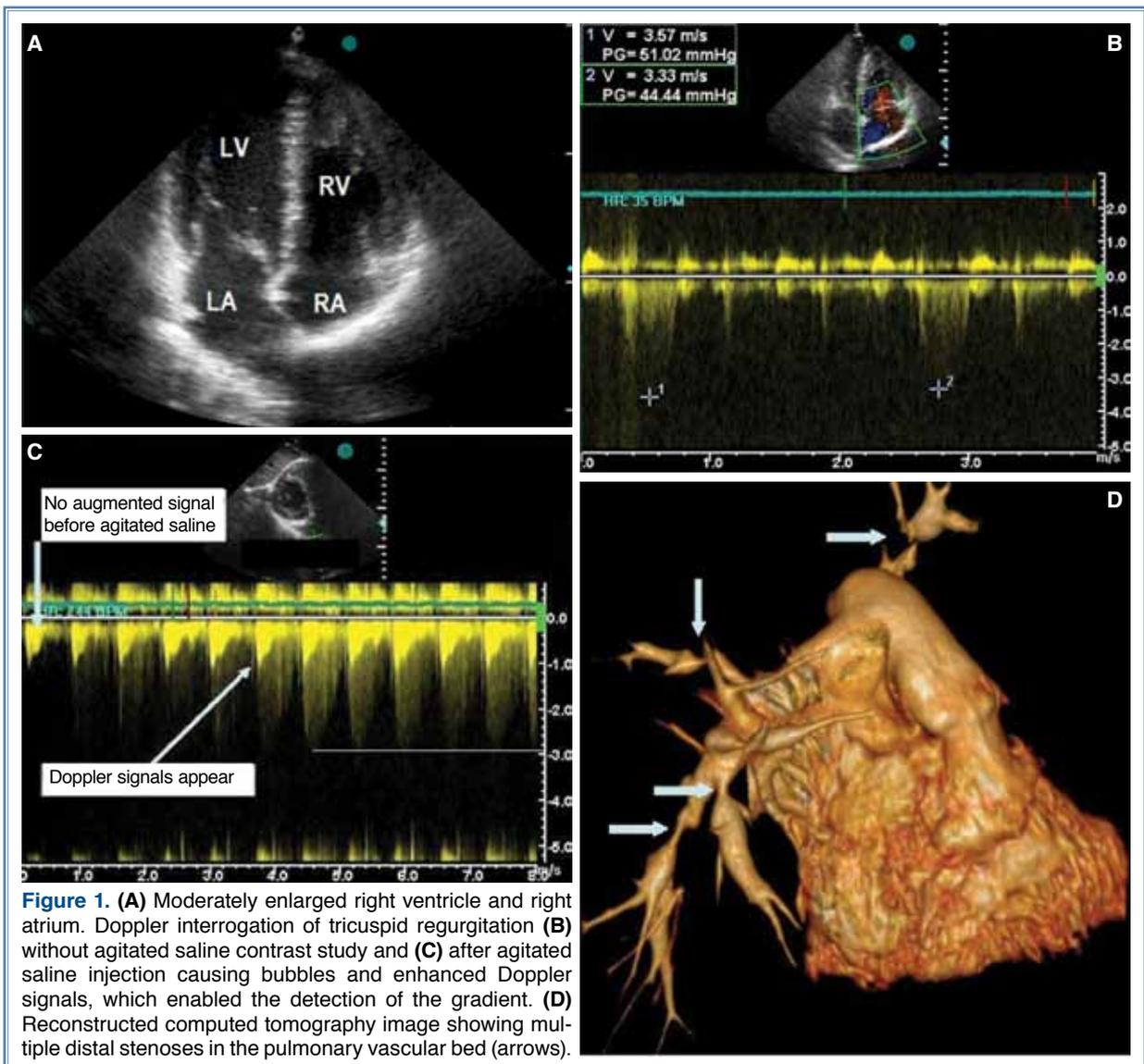
Correspondence: Dr. Zafer Işılak. GATA Haydarpaşa Eğitim Hastanesi, Kardiyoloji Servisi, 34668 Üsküdar, İstanbul, Turkey. Tel: +90 216 - 542 34 85 e-mail: drzaferisilak@gmail.com

© 2011 Turkish Society of Cardiology

CASE REPORT

A 19-year-old male patient was admitted to our outpatient clinic for examination of suitability for military service. History revealed exertional intolerance since early childhood. In a previous examination, he was told that he had a hole in his heart, which was not further analyzed. On physical examination, a murmur was heard over the entire right hemithorax. The patient was referred to the echocardiography unit with an initial diagnosis of ventricular septal defect. Echocardiographic examination showed that the left side of the heart was anatomically and functionally normal. The right ventricle, right atrium, and main pulmonary artery were moderately enlarged (Fig. 1a). Color Doppler echocardiography showed mild tricuspid regurgitation and no

flow across the interventricular septum. Right ventricular systolic pressure was estimated as 55-60 mmHg from the tricuspid regurgitation jet (Fig. 1b), which was later confirmed with agitated saline contrast echocardiography. Echocardiographic findings were consistent with pulmonary hypertension, probably of idiopathic or thromboembolic origin. However, this diagnosis did not account for the systolic murmur. In order to further evaluate the interventricular septum, agitated saline contrast echocardiography was performed for the presence or absence of contrast in the right ventricle or right-to-left shunt. The result was negative. However, during continuous wave Doppler examination of the right ventricular outflow tract while there were remnants of bubbles in the right heart and pulmonary vascular bed, a systolodiastolic flow with a peak gra-



dient of 30 mmHg was incidentally noted. The signal intensity was better when the cursor was aligned rightwards. After a few cycles that cleared the bubbles, the signal was not detectable. Repeat examination with agitated saline contrast again showed a gradient of 35 mmHg (Fig. 1c). When this finding was integrated with those of physical examination, a stenosis in the distal branches of the right pulmonary artery was suspected. However, the exact localization of the stenosis could not be determined by echocardiography and the patient was referred to computed tomography, which revealed multiple stenoses in the pulmonary vascular bed (Fig. 1d). The patient was informed about the disease and potential treatment modalities and was advised to consult a medical center where expenses of treatment could be covered by his insurance.

DISCUSSION

Distal pulmonary artery stenosis is a rare condition which frequently coexists with other congenital heart defects such as tetralogy of Fallot and arterial tortuosity syndrome, or a rubella infection during pregnancy. It may also present after arterial switch operations.^[2,3] This type develops in the neopulmonary artery. It is frequently overlooked and treated as idiopathic or thromboembolic pulmonary hypertension,^[4] but these stenoses do not respond to these treatments. In selected cases, stent implantation may improve the symptoms and echocardiographic findings.^[5] However, stent implantation is associated with high risk in patients with postoperative pulmonary artery stenosis and tetralogy of Fallot. In such patients, a collaborative transcatheter and surgical approach is needed.^[6] In our case, the patient did not receive any treatment until presentation to our clinic. Due to the presence of multiple stenoses in many distal branches, he was referred to a highly equipped medical center.

A careful examination is necessary for identifying patients with insidious onset of dyspnea and fatigue. The stenosis may sometimes be so distant and so small that the signals could not be detected by routine echocardiographic examination. In order to intensify the signals, contrast echocardiography may be used. Saline agitated with two syringes and a three-way stopcock is an easy method for making microbubbles. These microbubbles intensify the echodensity without affecting blood velocity and can enable the demonstration of blood flow with low-intensity Doppler signals. In our case, a signal detected by chance after saline contrast echocardiography enabled the diagnosis. At this point, it should be kept in

mind that agitated saline may increase the signals to the extent that they may sometimes be confused with pulmonary stenosis. The echocardiographer should notice whether the gradient is both systolic and diastolic for the final conclusion of a peripheral pulmonary stenosis, as is the case in aortic coarctation. The presence of only systolic gradient may be a result of agitated saline administration.

In conclusion, agitated saline contrast echocardiography should be used in all patients with anticipated right heart conditions. Its use should not be limited to estimation of pulmonary artery pressure and detection of patent foramen ovale. The pulmonary vascular bed should also be assessed by using agitated saline contrast echocardiography. We recommend the use of Doppler interrogation of the pulmonary vascular bed after agitated saline contrast injection in patients with increased right ventricular systolic pressure, especially in those without any explanatory concomitant disease. The interrogation should be in continuous Doppler mode with the depth adjusted to maximum as much as possible. Because the location of stenosis is not known, the ultrasonic beam must scan all range in multiple views.

Conflict-of-interest issues regarding the authorship or article: None declared

REFERENCES

1. Tokushima T, Utsunomiya T, Yoshida K, Ogawa T, Kido K, Ohtsubo Y, et al. Estimation of the systolic pulmonary arterial pressure using contrast-enhanced continuous-wave Doppler in patients with trivial tricuspid regurgitation. *Jpn Heart J* 1999;40:311-20.
2. Kalko Y, Hökenek F, Sever K, Demir T, Tireli E, Dayıoğlu E, et al. Arteriyel switch ameliyatı sonrası neo-pulmoner arter stenozu. *Türk Göğüs Kalp Damar Cer Derg* 2000;8:694-6.
3. Erdem A, Erol N, Zeybek C, Çelebi A. Arterial tortuosity syndrome in two cases. [Article in Turkish] *Türk Kardiyol Dern Arş* 2010;38:576-9.
4. Kreutzer J, Landzberg MJ, Preminger TJ, Mandell VS, Treves ST, Reid LM, et al. Isolated peripheral pulmonary artery stenoses in the adult. *Circulation* 1996;93:1417-23.
5. Saygılı A, Canter B, Kula S, Tunaoğlu FS, Olguntürk R. Stent implantation to left pulmonary artery stenosis in children: a case report. *Anadolu Kardiyol Derg* 2004;4:262-3.
6. Bacha EA, Kreutzer J. Comprehensive management of branch pulmonary artery stenosis. *J Interv Cardiol* 2001; 14:367-75.

Key words: Arterial occlusive diseases; contrast media; echocardiography; pulmonary artery/pathology/ultrasonography.

Anahtar sözcükler: Arter tıkaçıcı hastalık; kontrast maddesi; eko-kardiyografi; pulmoner arter/patoloji/ultrasonografi.