Atrial septal stenting to increase interatrial shunting in cyanotic congenital heart diseases: a report of two cases

Siyanotik doğuştan kalp hastalıklarında interatriyal şantı artırmak amacıyla atriyal septuma stent uygulaması: İki olgu sunumu

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Summary – Aiming to increase mixing at the atrial level, atrial septal stenting was performed in two pediatric cases with cyanotic congenital cardiac diseases. The first case was a 3-month-old male infant with transposition of the great arteries. The second case was an 18-month-old male infant with increased central venous pressure due to postoperative right ventricular outflow tract obstruction. Premounted bare stents of 8 mm in diameter were used in both cases. The length of the stent was 20 mm in the first case and 30 mm in the latter. The procedure was accomplished without any complications. In the first case, oxygen saturation increased approximately 20-25% with no significant interatrial gradient. In the latter, central venous pressure decreased from 16 to 8 mmHg immediately after the procedure. The patient was weaned from the ventilator on the second day and discharged from intensive care unit on the fifth day. Follow-up echocardiograms of both patients showed patent stents with good position relative to the atrial septum. Stenting of the atrial septum seems to be a safe and effective method to create a reliable, nonrestrictive interatrial communication.

Özet – Siyanotik doğuştan kalp hastalığı tanısıyla izlenen iki bebekte, atriyal düzeyde karışımı artırmak amacıyla atriyal septuma stent yerleştirme işlemi uygulandı. Birinci olgu, büyük arterlerin transpozisyonu tanısıyla izlenen üç aylık bir erkek bebekti. Diğer olgu, ameliyat sonrası dönemde sağ ventrikül çıkım yolu tıkanıklığına bağlı olarak santral venöz basınç yüksekliği gelişen 18 aylık bir erkek bebekti. Her iki olguda da 8 mm çapında, balona monte edilmiş çıplak stent kullanıldı. Stent uzunluğu ilk olguda 20 mm, ikinci olguda 30 mm idi. İşlem herhangi bir komplikasyon olmadan gerçekleştirildi. İlk olguda oksijen satürasyonu işlem sonrasında yaklaşık %20-25 yükseldi ve interatrial gradiyent kalmadı. İkinci olguda, santral venöz basınç işlemden hemen sonra 16 mmHg'den 8 mmHg'ye düştü. Hasta işlemin ikinci gününde ventilatörden ayrıldı ve beşinci günde yoğun bakım ünitesinden taburcu edildi. Kontrol ekokardiyografik incelemelerde, her iki olguda da stentin açık ve atriyal septumda iyi pozisyonda olduğu görüldü. Atriyal septuma stent yerleştirilmesi, güvenilir ve açık bir interatriyal bağlantı yaratmada güvenli ve etkili bir işlem olarak görünmektedir.

Interatrial shunting through an atrial septal defect is vital in certain conditions in pediatric cardiology. Transposition physiology and left- or right-sided obstructive lesions are the major examples that need sufficient blood flow through the atrial septal defect, so creating or enlarging an interatrial communication may be required in these pathologies.[1,2]

Balloon atrial septostomy in patients with transposition of the great arteries is highly successful in the first several weeks of life.[3] However, beyond the neonatal period, the atrial septum gets thicker and becomes vulnerable to tears during balloon septostomy. Blade septostomy has been introduced in cases in which balloon septostomy fails, but this procedure

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presents significant risks.[4] Static and cutting balloons have also been used to enlarge interatrial communications, also with uncertain results.[5] Recently, stenting of the atrial septum has been introduced to achieve a reliable interatrial communication.[6,7]

We report on atrial septal stenting procedures to create a nonrestrictive atrial communication in two patients, one with transposition physiology, and the other with postoperative increase in central venous pressure related to right ventricular inflow and outflow obstruction.

CASE REPORT

Case 1– A 3-month-old male infant with the diagnoses of transposition of the great arteries, restrictive ventricular septal defect, pulmonary valvular stenosis, and significantly restrictive atrial communication was admitted with frank cyanosis. He was cyanotic (oxygen saturation low 50's) and had mild respiratory distress. His heart sounds were normal and there was a 3/6 systolic murmur at the upper left sternal border. There was no organomegaly. Peripheral pulses were equally palpable in all four extremities. Echocardiographic examination revealed levocardia with atrial situs solitus, atrioventricular concordance with ventriculoarterial discordance, a very small (1 mm?) atrial communication (Fig. 1a) with a minimal left-to-right shunt, and a restrictive outlet ventricular septal defect (3.5 mm). The pulmonary valve annulus was mildly hypoplastic (Z-score: -2) and the valve was stenotic. The peak systolic pressure gradient across the pulmonary valve was at least 60 mmHg by continuous-wave Doppler interrogation.

With these findings, the patient was taken to the cardiac catheterization laboratory for urgent balloon atrial septostomy. Since the atrial communication was very small, it was somewhat difficult to cross the septum. Thus, a 0.014" coronary guide wire was advanced across the septum and predilation was done using a coronary balloon catheter (5 x 20 mm). Then, we were able to perform balloon atrial septostomy with limited success (Fig. 1b). Static atrial balloon septoplasty was then performed using a 10 mm x 2 cm followed by a 14 mm x 2 cm Tyshak balloon valvuloplasty catheter (NuMed Inc, Ontario, Canada). An atrial communication about 5 mm in size was finally achieved. There was still a 3-4 mmHg pressure gradient between the two atria. Oxygen saturation increased to high 60's to low 70's immediately after the procedure and the patient was transferred to intensive care unit for follow-up. However, oxygen saturation gradually dropped to low 60's within two days of hospital stay and, echocardiographically, the size of the atrial communication decreased to 2-3 mm, suggesting that the septum had not actually been teared but only stretched during the initial intervention. Thus, we planned to stent the atrial septal defect in order to achieve a nonrestrictive interatrial communication. The patient was taken to the catheterization laboratory for stenting the atrial septum.

General anesthesia was administered and central venous access was obtained via the femoral vein. The patient underwent diagnostic right and left heart catheterization. Access to the left atrium was obtained via the restrictive interatrial communication. A wire was positioned in the upper right pulmonary vein after looped in the left atrium, and an 8 Fr guiding catheter was advanced over the wire, with the tip across the atrial septum. The catheter preloaded by an 8 x 20-mm cobalt iliac stent (Assurant, Medtronic, Minneapolis, USA) was advanced through the guiding catheter and across

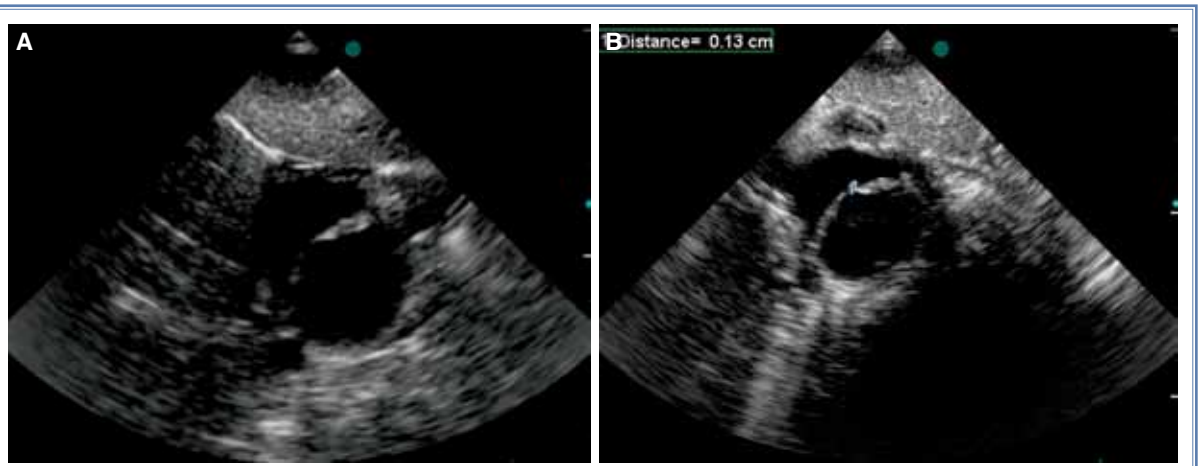

Figure 1. Echocardiographic demonstration of a highly restrictive interatrial communication **(A)** before and **(B)** after balloon atrial septostomy (Case 1).

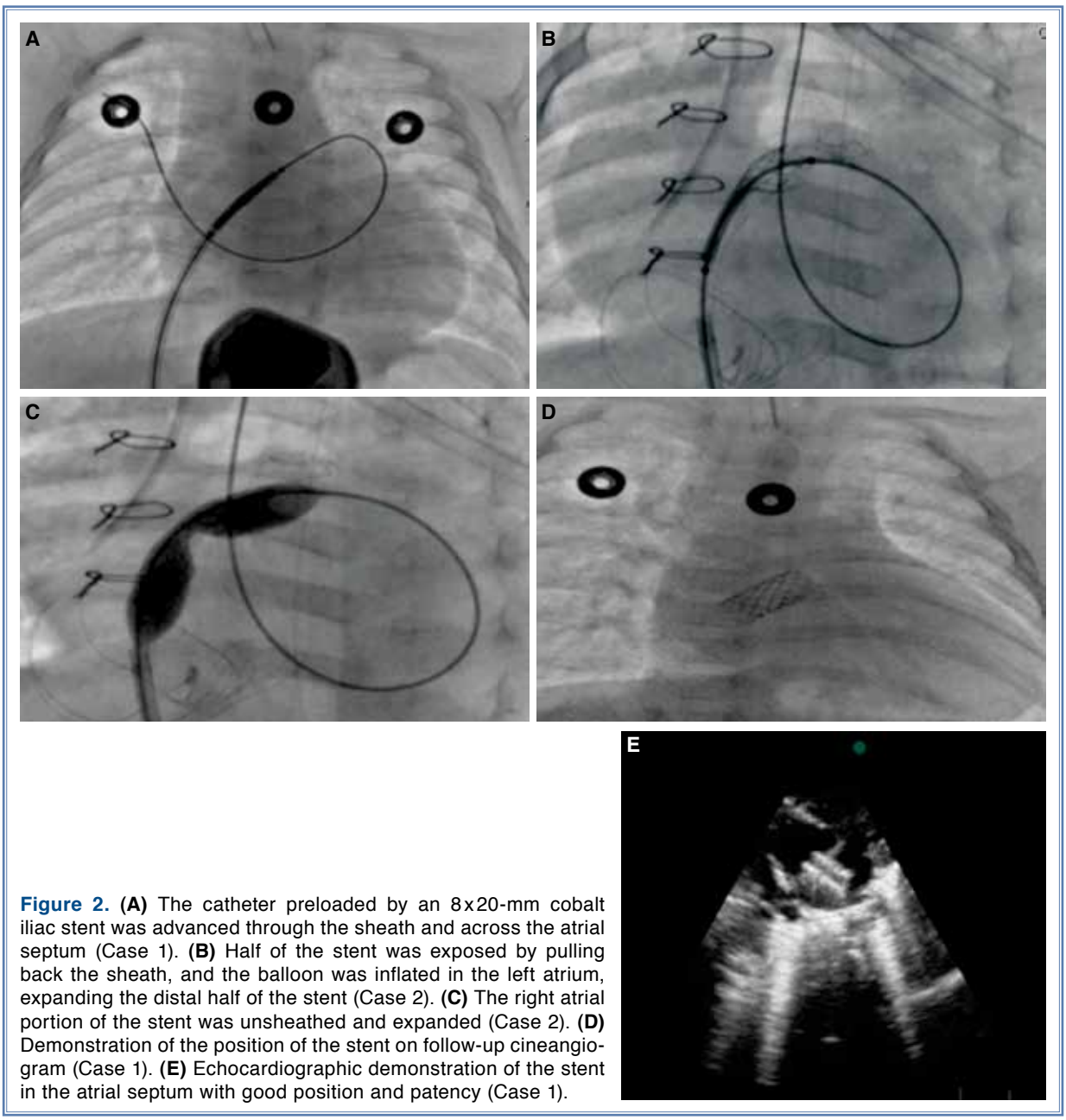

Figure 2. **(A)** The catheter preloaded by an 8x20-mm cobalt iliac stent was advanced through the sheath and across the atrial septum (Case 1). **(B)** Half of the stent was exposed by pulling back the sheath, and the balloon was inflated in the left atrium, expanding the distal half of the stent (Case 2). **(C)** The right atrial portion of the stent was unsheathed and expanded (Case 2). **(D)** Demonstration of the position of the stent on follow-up cineangiogram (Case 1). **(E)** Echocardiographic demonstration of the stent in the atrial septum with good position and patency (Case 1).

the atrial septum (Fig. 2a). Half of the stent was exposed by pulling back the guiding catheter, and the balloon was inflated in the left atrium, expanding the distal half of the stent (Fig. 2b, Case 2). Next, the entire system was firmly pulled back against the atrial septum, and the right atrial portion of the stent was unsheathed and expanded (Fig. 2c, Case 2). The position of the stent was confirmed by a cineangiogram (Figure 2d). After stent implantation, there was no significant pressure gradient between the two atria and oxygen saturation increased to mid 80's. Echocardiography showed good stent position (Fig. 2e). During a follow-up of two months, there was no change in oxygen saturation level and the patient was thriving well.

Case 2– An 18-month-old male patient was admitted with the complaints of cyanosis and failure to thrive. He was cyanotic (oxygen saturation low 60's), mildly dyspneic, and had tachycardia. There was a faint continuous murmur at the upper left sternal border. The liver was palpable 2 cm below the costal margin. Echocardiography revealed pulmonary atresia and a moderately restrictive outlet ventricular septal defect with suprasystemic right ventricular pressure. The right ventricle and tricuspid valve were mildly

hypoplastic. The pulmonary arteries were confluent but hypoplastic, principally fed by a ductus arteriosus. The size of the branch pulmonary arteries was found inadequate for total correction. To augment pulmonary arteries and pulmonary flow, a central aortic-to-pulmonary artery shunt was performed. The patient's postoperative course was complicated with prolonged endotracheal intubation with increased central venous pressure (16-18 mmHg), hepatomegaly (4 cm below the costal margin), and pleural effusions.

Postoperative echocardiographic examinations showed significant restriction at the interatrial communication and a decision was made to create a nonrestrictive right-to-left shunt to decrease central venous pressure. Because of the patient's unstable condition, a transcatheter approach was planned. As the chance of a successful balloon atrial septostomy at 18 months of age was very low and static septoplasty might fail to provide a long-lasting nonrestrictive communication, we planned to stent the atrial septum. The patient was taken to the catheterization laboratory and an 8 x 30-mm premounted cobalt iliac stent (Assurant, Medtronic) was implanted following the same procedure previously described in the first case. After stent implantation, central venous pressure decreased to 8 mmHg immediately. Hepatomegaly gradually decreased. The patient was weaned from the ventilator on the second postprocedural day. His clinical condition got better and he was discharged from intensive care unit on the seventh day of stenting.

DISCUSSION

A stent is an artificial tube inserted into a natural passage or conduit in the body to prevent or counteract a congenital or acquired flow restriction. It is a mesh-like tube of thin wire. Stents may be bare metal or covered with an outer polytetrafluoroethylene membrane, self-expanding or balloon-expandable, and hand-mounted on the delivery balloon or premounted.[8]

Since the first implantation of a spiral coil-spring prosthesis in experimentally created vascular stenotic lesions, endovascular and intracardiac stenting has undergone a tremendous improvement. In the early 90's, stents were started to be used in congenital cardiac stenotic lesions.[8,9] Stenting is now the standard and first-line treatment for coarctation of the aorta in adults or postoperative pulmonary artery branch stenoses.[8]

Stents have been used not only in stenotic lesions, but also to ensure patency of naturally occurring or artificially created intercirculatory communications.[6-8] In this report, we described our experience with atrial septal stenting to enlarge interatrial communications in two patients, one with transposition physiology, and the other with postoperative right heart failure and elevated central venous pressure. Indications were clear and the procedure was life-saving in both cases. Other potential indications of atrial stenting include hypoplastic left heart syndrome to decrease left heart pressures, pulmonary hypertension with right ventricular failure, and atrioventricular valvular atresia or hypoplasia to decrease pulmonary or systemic venous pressures.[7,8,10]

The oldest and the most widely used material for balloon-expandable stents is stainless steel. Alternative materials are cobalt alloys, platinum alloys, and tantalum. Newly developed cobalt-chromium alloy stents have lower crimping profiles with high radial strength.[8] In most cases of atrial stenting, premounted stents were used, ranging from 7 to 9 mm in diameter and from 12 to 26 mm in length.[6-8] We used FDA-approved premounted bare stents made from cobalt in both cases. Stent diameter was 8 mm in both cases. Stent length was 20 mm in the first patient weighing 5 kg, and 30 mm in the other patient weighing 10 kg. Echocardiographically measured biatrial diameter was at least 1.5 to 2 times the length of the stent. Careful manipulation under both echocardiographic and angiographic guidance was done to avoid close contact of the stent with the atrial free walls or nearby cardiac structures.

Potential complications of the procedure are stent migration, stent malposition, stent fracture and dissection, injury to the neighboring structures, thrombosis, bleeding, and even death in young infants. Stent migration or malposition may occur in cases with an aneurysmatic septum, or the stretch in the atrial septum may not be sufficient for the stent to stay in its desired location. Therefore, the operator must be very careful not to leave stent loose in its position while inflating the balloon, and not to oversize stent that would result in tearing the septum.

Dissection and injury to the neighboring structures may be closely related to the stent length and malposition. In young infants, the atrial length is important for the stent to prevent injury to cardiac structures. It is important to measure the total biatrial length and left and right atrial lengths separately in case of atrial hypoplasia. It may be necessary to implant less than half of the stent in the hypoplas-

tic site in order to avoid contact to the atrial wall or nearby structures.

One of the most feared complications of atrial stenting is thrombus formation. Although some authors use oral anticoagulants for prevention of potentially lethal thrombotic stent occlusion, most of the clinicians use aspirin alone at antithrombotic doses.[8,10] We used intravenous heparin for 24 hours and continued with oral aspirin at antithrombotic doses.

No serious complications occurred in our cases related to the procedure. The immediate and short-term results of the procedure were excellent. There was a significant improvement in oxygen saturation in the first patient, and a significant decline in central venous pressure in the other. The first patient was discharged on the second day with an oxygen saturation of approximately 85%. The second patient was weaned from the ventilator and discharged from intensive care unit after a week. Follow-up echocardiographic examinations in the first and third months demonstrated excellent persistence of the atrial communication in both patients, with no evidence for stent thrombosis or malposition.

In our opinion, atrial septal stenting seems to provide a long-lasting, nonrestrictive atrial communication and should be taken into consideration as an alternative to surgery as well as other interventional procedures including blade atrial septostomy, in cases where conventional treatment of balloon atrial septostomy, static or cutting balloon dilatation of the atrial septum fail. Our limited experience suggests that the procedure is reliable, safe, and effective. However, the stents used for the procedure are not designed specifically for this purpose and may not be ideal in young infants and children. The device that meets the requirements of an ideal stent to be used for atrial septum stenting in infants and young children has yet to be developed.

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Key words: Balloon dilation; cyanosis/etiology; heart catheterization; heart septal defects, atrial/therapy; heart septum; infant; stents.

Anahtar sözcükler: Balonla genişletme; siyanoz/etyoloji; kalp kateterizasyonu; kalp septal defekti, atriyal/tedavi; kalp septumu; bebek; stent.