aorta includes coronary artery disease. Coronary artery disease seems to be accelerated in these patients (5). Although the etiology of this accelerated atherosclerotic heart disease is unknown, a number of factors probably play a role. Hypertension is a known risk factor for the development of coronary artery disease (6). There must be factors other than hypertension, because even considering the higher incidence of hypertension, the incidence of coronary artery disease is much higher than one would expect. Some individuals with coarctation of the aorta are known to have altered structure and function of both the coronary and other systemic arteries. Chen et al. (7) described severe atherosclerosis and calcification in internal mammary arteries of two patients with previous coarctation repair who required coronary artery bypass surgery (7). In clinical practice, the development of dissection in a patient with coronary artery disease is common immediately after balloon angioplasty because of intimal tearing. No spontaneous coronary dissection in a patient with coarctation of aorta was reported in literature.

Conclusion

Early coronary lesions should be sought in the follow-up of patients with native aortic coarctation.

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Intractable atrial flutter presented with severe bradycardia in an infant

Süt çocukluğu döneminde ciddi bradikardi ile ortaya çıkan tedaviye yanıtsız atriyal flatter olgusu

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Introduction

Atrial flutter is an uncommon arrhythmia in newborns and infants. Generally, it leads to tachycardia and is not associated with congenital heart disease in neonatal period. In most cases, it is easily converted to sinus rhythm by medical or electrical cardioversion. After resolution of the arrhythmia, the long-term prognosis is excellent. However, some patients may have intractable atrial flutter despite to the treatment.

We report a 3 months of age infant who presented with severe bradycardia and diagnosed as atrial flutter without response to treatment that has not previously been reported.

Case Report

The 3 months of age infant with bilateral congenital cataract was referred to our department because of bradycardia. In physical examination, the heart rate was 40 beats per minute (bpm), respiratory rate was 60 per minute, blood pressure 85/60 mmHg and bilateral cataract formation was detected. Electrocardiography (ECG) revealed bradycardia with a ventricular rate of 57 bpm and intraventricular conduction block (QRS: 0.12 s) (Fig. 1). Atrial flutter waves were not detected on the 12-lead ECG. Mild cardiomegaly was observed on chest X-Ray (cardiothoracic ratio, 0.56). The echocardiogram revealed a

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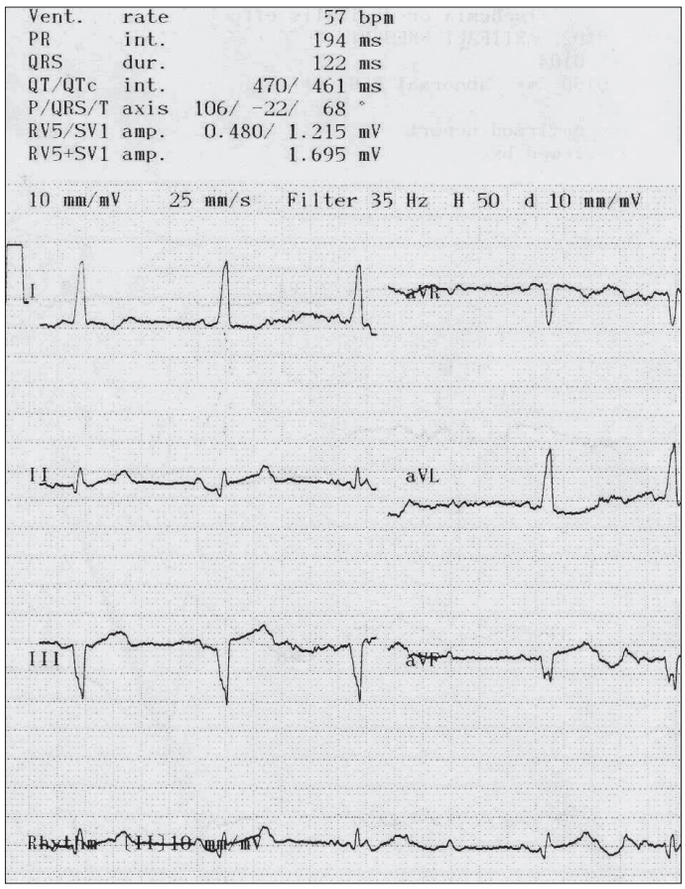

Figure 1. The electrocardiogram shows bradycardia with a ventricular rate of 57 bpm and intraventricular conduction block (QRS: 0.12 s)

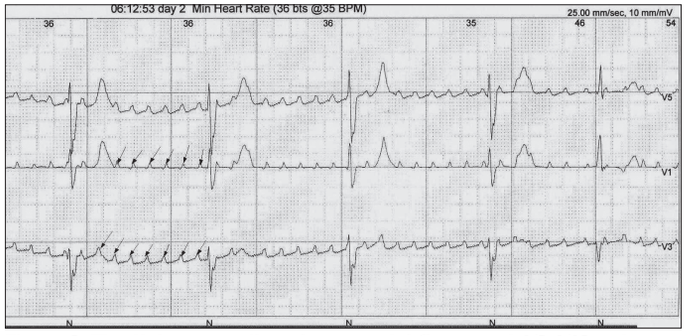

Figure 2. The Holter recording shows atrial flutter and 1/8-1/10 atrioventricular block with a ventricular rate of 36 bpm

structurally normal heart and a small amount of pericardial fluid. The 24 h Holter recording revealed atrial flutter with an atrial rate of 320 bpm and 1/8-1/10 atrioventricular block with a ventricular rate of 36 bpm (Fig. 2). The etiology of the cataract was investigated. Urine organic acids, blood amino acid chromatography analysis, and genetic karyotype were normal. Infectious analysis (toxoplasmosis, rubella, cytomegalovirus, and herpes simplex [TORCH]) were negative. Because of severe bradycardia, transcatheter temporary ventricular pacing was applied and then direct current (DC) cardioversion was performed, which failed. Next, antiarrhythmic drugs, including propafenone (1 mg/kg) intravenously, sotalol (100-200 mg/m^2 /day) per oral for 2 days, and amiodarone (5 mg/kg) intravenously were tried during transcatheter temporary ventricular pacing. These drugs, however, also had no effect. After then, permanent epicardial pacemaker was implanted. During follow-up despite amiodarone treatment, atrial flutter was not converted to sinus rhythm. The patient with permanent epicardial pacemaker was followed-up 8 months without any complication.

Discussion

Atrial flutter in infants is a well-tolerated arrhythmia and generally not associated with congenital heart disease. However, patients with transposition of the great arteries or single ventricle who have undergone Mustard, Senning, or Fontan procedures are more prone to develop this arrhythmia because of atrial scars from surgery and right atrial enlargement (1, 2). The diagnosis of atrial flutter generally can be made from ECG, which most frequently shows 2:1 atrioventricular conduction, but if atrioventricular conduction is impaired, there can be 3:1 or 4:1 atrioventricular conduction. Atrial flutter generally leads to tachycardia. The potential exists for development of congestive heart failure, which is appeared to be related to the duration of the tachycardia (3). Texter at al. (1) showed 10 infants who presented with symptoms of congestive heart failure, all had a history compatible with prolonged tachycardia. Atrial flutter rarely leads to bradycardia because of high-grade atrioventricular block, similar to our case.

The severity and prognosis of arrhythmia are evaluated via transesophageal electrophysiologic studies more accurately. These are reliable in assessment of the function of the sinus node, anterograde atrioventricular conduction, refractory period of atria, and atrioventricular accessory connections, which can provoke and terminate tachycardia, and the efficacy of antiarrhythmic drugs. However, in infants this is not routinely used because of technical difficulty (4).

Infants with atrial flutter have a good prognosis and convert to sinus rhythm spontaneously, or by DC cardioversion or transesophageal overdrive pacing. Direct current cardioversion and transesophageal overdrive pacing are safe methods to attempt restoration of sinus rhythm. However, DC cardioversion appeared more successful than transesophageal overdrive pacing at establishing sinus rhythm. Chronic antiarrhythmic therapy is not necessary, however; atrial flutter associated with postoperative procedure may be resistance to electrical cardioversion or persistent, requiring the use of antiarrhythmic medications (1, 5). Digoxin, quinidine, propranolol, sotalol, flecainide, propafenone and amiodarone have also been used with varying degrees of success (1, 5-6). Seventy-two cases of neonatal or infantile atrial flutter have been reported in the literature (6). In these cases, 6 patients recovered sinus rhythm spontaneously, 16 recovered it following digoxin administration, 2 recovered it following combined digoxin and sotalol therapy, 1 recovered after flecainide administration, 42 recovered it following electrical cardioversion (DC cardioversion or atrial pacing). Five other patients did not respond to therapy. Despite of propafenone, sotalol and amiodarone treatment, our patient was not converted to sinus rhythm.

Radiofrequency catheter ablation is a successful and effective cure for a variety of supraventricular tachycardias. In infants and young children, there is a high rate of spontaneous resolution of their tachyarrhythmias after the first year of life, for these reasons, the indication for radiofrequency ablation in children younger than 4 years should be more restrictive compared to older children and adolescent patients (4, 7).

Atrioventricular block can be divided into two types: congenital or acquired. Congenital complete atrioventricular block may be due to

mothers' collagen vascular disease such as systemic lupus erythematosis, discoid lupus, Sjogren syndrome, etc. Many of the mothers were diagnosed as a result of finding of congenital atrioventricular block in the baby (8). The antibodies responsible for this passive transplacental autoimmune injury were shown to be SSA/Ro and SSB/ La. Other etiologies of congenital complete atrioventricular block include myocarditis, tumors and, patients with long QT syndrome. Acquired complete atrioventricular block is most associated with surgical correction of a congenital heart defect. This includes ventricular septal defects, either as isolated defects or as part of a more complex congenital heart defect, i.e., tetralogy of Fallot, atrioventricular septal defects, or L-transposition of great arteries with ventricular inversion. In our case, atrial flutter with an atrial rate of 320 bpm and 1/8-1/10 atrioventricular block with a ventricular rate of 36 bpm was detected by 24 hours Holter recording. The indications for permanent pacing in pediatric patients have been recently revised (9). Permanent pacemaker implantation is indicated for congenital third degree atrioventricular block in the infant with a ventricular rate less than 55 bpm or with congenital heart disease and a ventricular rate less than 70 bpm. Our patient was asymptomatic and had not congenital heart disease but had severe bradycardia. Therefore, permanent epicardial pacemaker was implanted because of increased risk of sudden death.

Conclusion

In conclusion, atrial flutter is a rare tachycardia in the newborn and young infant and usually has an excellent prognosis. Persistent bradycardia may be due to atrial flutter with high atrioventricular bloke. Because of this reason, cases with bradycardia should be awareness as differential diagnosis with atrial flutter.

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