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CASE REPORT

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Efficacy and safety of cardioversion with continuous landiolol infusion for atrial tachyarrhythmia in an inflammatory state caused by volvulus in a child with TARP syndrome and postoperative tetralogy of Fallot

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Abstract

A 2-year-old boy was diagnosed with TARP syndrome and underwent surgery for tetralogy of Fallot. He developed fever and had an acute abdomen. After 12 hours, atrial tachyarrhythmia (300 beats/min [bpm]) occurred. After nine administration of adenosine and two cardioversions, it relapsed promptly. Landiolol (10 μg/kg/min) was administered until the heart rate decreased to 270 bpm, and cardioversion was performed until sinus rhythm was normal. Exploratory laparotomy revealed small bowel volvulus. Systemic inflammation causing an acute abdomen may be associated with atrial tachyarrhythmia in postoperative tetralogy of Fallot. We speculated that landiolol lowered the defibrillation threshold of the atrium.

KEYWORDS
β-blocker, atrial tachyarrhythmia, Landiolol, laparotomy, TARP

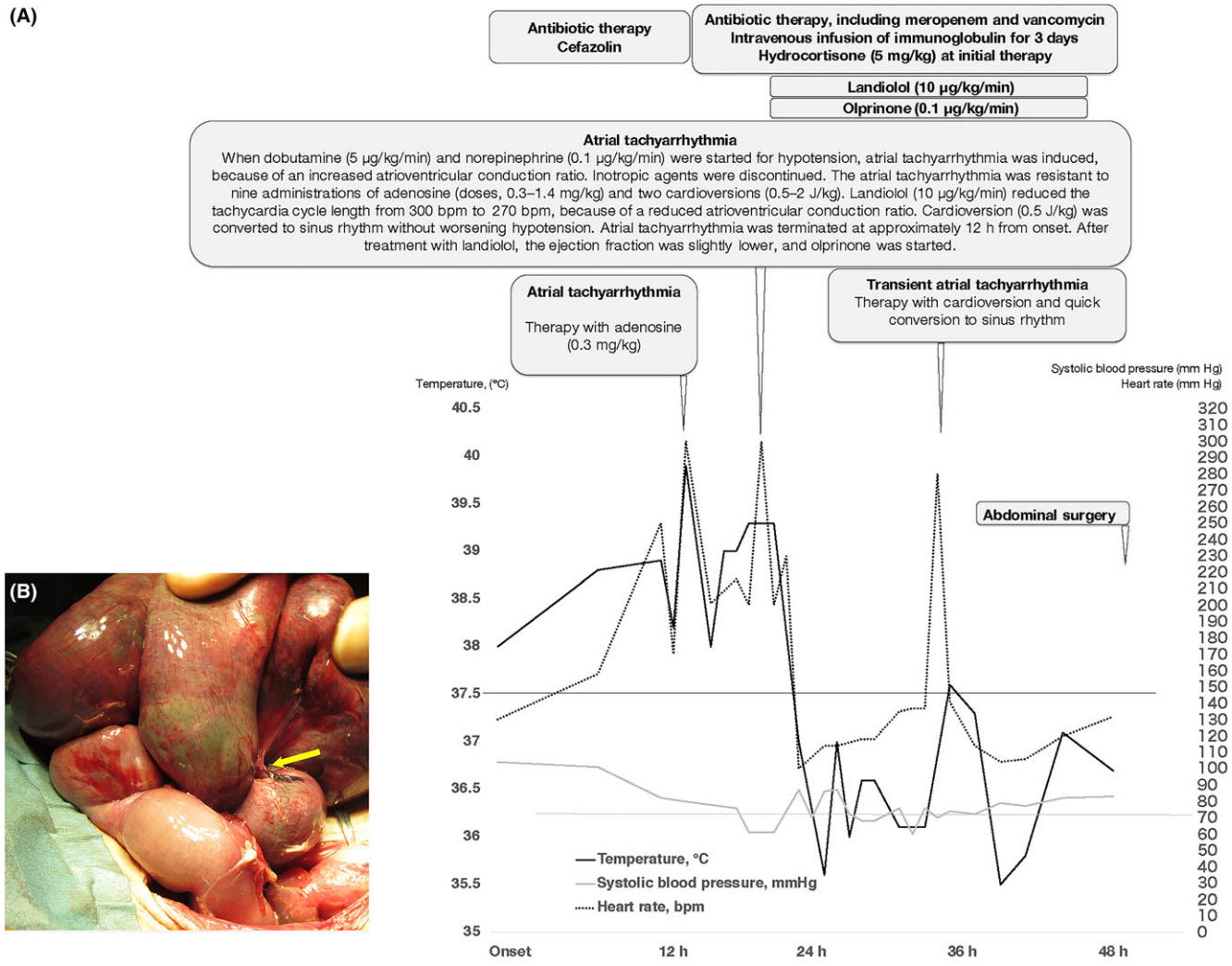
1 | INTRODUCTION

Although landiolol, an ultrashort-acting β-blocker, is effective for atrial tachyarrhythmia in adult patients, its efficacy in pediatric patients is unclear.[1,2] Here, we report the effectiveness of landiolol for atrial tachyarrhythmia in a 2-year-old boy with tetralogy of Fallot and TARP syndrome.

2 | CASE REPORT

A 2-year-old boy was diagnosed with tetralogy of Fallot and a persistent left superior vena cava. He presented with talipes equinovarus, Pierre Robin sequence, and heart disease. Genetic testing revealed an *RBM10* gene mutation with TARP syndrome.[3] He underwent intracardiac repair and tracheotomy for tracheal stenosis at 2 years of age. After surgery, captopril therapy for heart failure and tubal feeding of an elemental diet for oral intake difficulty were continued during hospitalization. He was hospitalized for a long time as his mother was concerned about the home medical care associated with feeding, management of the tracheotomy tube, and administration of drugs. She was a single mother and had no support from the rest of her family. The patient had a clubfoot, a high arched palate, low set ears, a small jaw, vermis hypoplasia, and hearing difficulty on both sides. Echocardiography showed a left ventricular ejection fraction of 53%, mild pulmonary stenosis, and regurgitation with right ventricle volume overload. Electrocardiography showed right axis

deviation and complete right bundle branch block. At 2 years of age (weight, 7.2 kg; height, 83 cm), he developed fever and abdominal distension one morning, and cefazolin was administered. In the night, he developed high-grade fever and tachycardia (Figure 1A). Because of suspected atrial tachyarrhythmia, adenosine was administered (0.3 mg/kg rapid injection), and his heart rate decreased from 300 to 200 beats/min (bpm) (Figure 2A). The diagnosis of atrial tachyarrhythmia remained. He had hypoglycemia (27 mg/dL), increased white blood cell count (12,900/μL), and an elevated C-reactive protein level (28 mg/dL). He showed abdominal distension with a dilated colon, and thus, acute abdomen was diagnosed. Hydrocortisone (5 mg/kg), meropenem, vancomycin, and intravenous immunoglobulin were administered. His body temperature and heart rate decreased slightly because of responses to the initial therapies, but his high-grade fever and tachycardia persisted. The next morning, on administration of inotropic agents to raise his lower systolic blood pressure (70 mm Hg, lower limit of systolic blood pressure was 74 mm Hg), a tachycardia cycle of 300 bpm was induced. The administration of inotropic agents was stopped, as they may cause an increase in the atrioventricular conduction ratio. The atrial tachyarrhythmia was resistant to nine administrations of adenosine (doses, 0.3-1.4 mg/kg) (the atrioventricular conduction ratio decreased within a few minutes) and two cardioversions (0.5-2 J/kg) (termination and conversion to sinus rhythm transiently). Treatment with landiolol (dose, 10 μg/kg/min) reduced the heart rate from 300 to 270 bpm (Figure 2B), with a



Blood test									
While blood cell count, /μl	14600			12900	6900	8100	11100	12900	9100
C-reactive protein level, mg/dl	3.63			28.01	15.33	17.35	25.14	28.89	29.85
B-type natriuretic peptide level, pg/ml						5656			
Ultrasonography									
LVDD, mm		**25**	**24**						**27**
Ejection fraction, %		**66**	**51**						**51**

FIGURE 1 A, Clinical course of our patient's acute abdomen and wide QRS tachycardia. B, Exploratory laparotomy shows a strangulated ileus. Small bowel volvulus with malrotation is diagnosed (the site of volvulus is shown with a yellow arrow). Excision of the small bowel is performed. LVDD, Left ventricular end-diastolic diameter

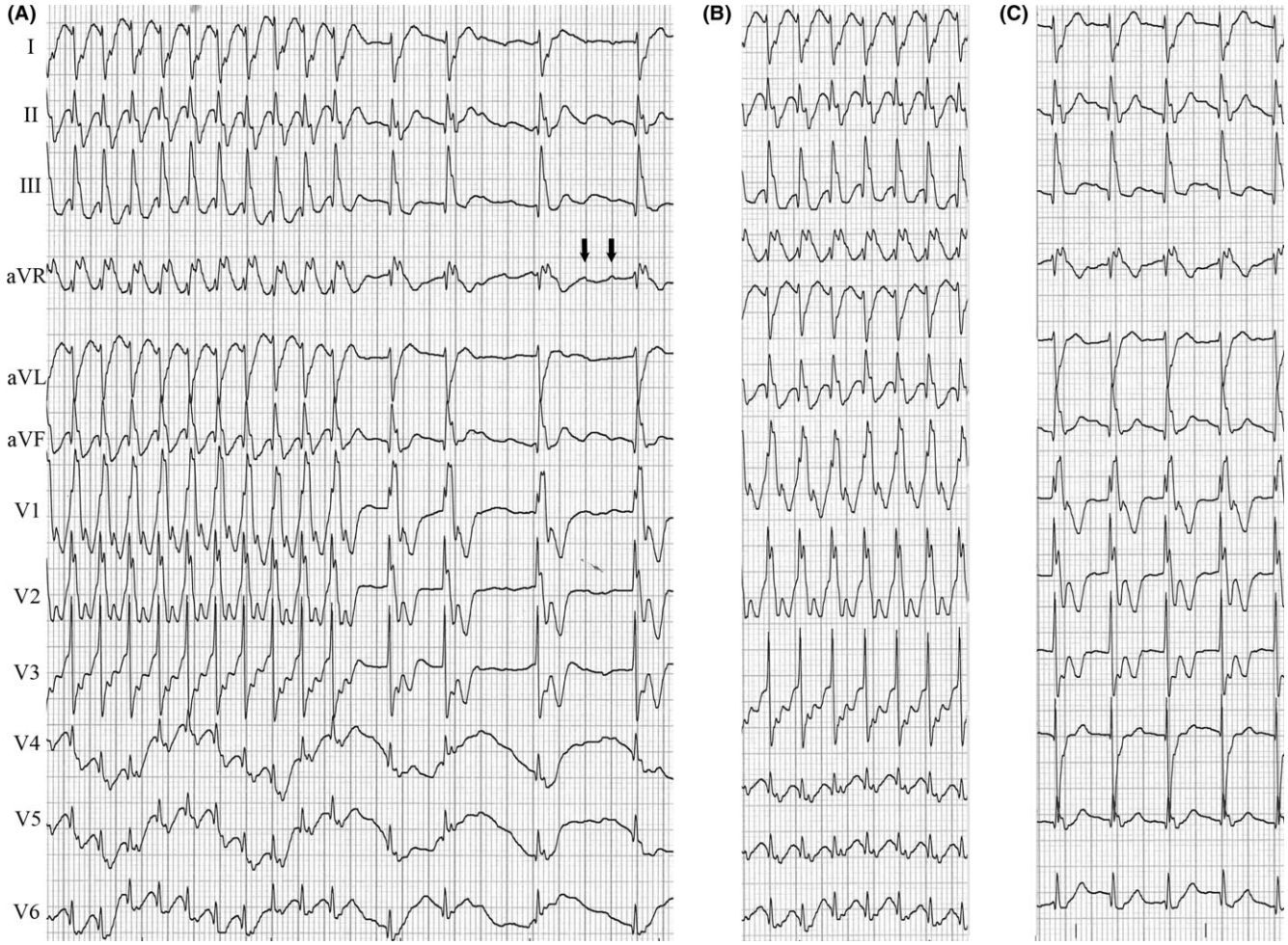


FIGURE 2 A, Complete right bundle branch block is complicated at sinus rhythm. The tachycardia rate is 300 beats/min. Atrial tachyarrhythmia is retrospectively diagnosed because of a continuous atrial wave at adenosine infusion (arrow). B, The heart rate with landiolol during the 20-min tachycardia cycle reduces from 300 to 270 beats/min. C, Conversion to sinus rhythm is noted after cardioversion with continuous landiolol infusion

reduced atrial rate and atrioventricular conduction ratio. Then, cardioversion of 0.5 J/kg initiated sinus rhythm without worsening of the low blood pressure (Figure 2C). Atrial tachyarrhythmia was terminated at approximately 12 hours from onset. The atrial tachyarrhythmia persisted until commencement of landiolol administration and cardioversion. His body temperature decreased from 39.3 to 36.3°C. Amiodarone was not used because of sinus bradycardia after cardiac surgery (40-50 bpm at rest). Echocardiography showed a lower ejection fraction of the left ventricle, and olprinone was started (Figure 2). Atrial tachyarrhythmia showed transient recurrence, and it was converted to sinus rhythm through treatment, including cardioversion and landiolol administration. After these therapies, exploratory laparotomy of his strangulated ileus was performed, and small bowel volvulus with malrotation was diagnosed. Surgical excision of the small bowel was performed (Figure 1B). We made a retrospective diagnosis of atrial tachyarrhythmia with an atrial wave at tachycardia on electrocardiography (Figure 2A). Landiolol was switched to bisoprolol after abdominal surgery.

3 | DISCUSSION

The clinical course of this patient highlights two important points. First, landiolol might lower the defibrillation threshold of the atrium. Second, the combination of cardioversion and landiolol administration is effective for atrial tachyarrhythmia in postoperative congenital heart disease.

The safety of landiolol has not been clearly established in children, and few reports have mentioned the safety and efficacy of landiolol in children.[4] A previous study showed that treatment with landiolol for electrical storm associated with increased sympathetic nerve activity in adults with myocardial infarction was effective, without hemodynamic collapse, after 100 electrical defibrillations.[5] Another study showed that landiolol was effective and safe for a class III antiarrhythmic drug-resistant electrical storm.[6] However, it is unclear whether a β-blocker is the most appropriate drug for atrial tachyarrhythmia in children. The response of an advanced electrical storm is evoked by enhanced sympathetic nerve activity. Additionally, β-blockers, particularly landiolol, are known to be effective for

suppressing sympathetic nerve activity. Moreover, the atrial rate and atrioventricular conduction ratio can be lowed to the levels at defibrillation of the atrium by reducing sympathetic nerve activity. Thus, we speculated that landiolol lowered the defibrillation threshold of the atrium and inhibited sympathetic nerve activity associated with inflammation.

Landiolol has been shown to be safe and effective for the treatment of atrial arrhythmia in adults.[1,2] This case showed that cardioversion did not cause suppression with stimulation of the arrhythmogenic substrates of atrial tachyarrhythmia. Only continuous landiolol infusion combined with cardioversion was effective for suppressing the substrate in our patient. During landiolol and cardioversion treatments for atrial tachyarrhythmia, our patient did not show collapse of circulatory dynamics, as reported previously.[5] A previous study showed that landiolol was effective for junctional ectopic tachycardia in pediatric patients with postoperative congenital heart disease.[4] However, the efficacy of landiolol for atrial tachyarrhythmia has been rarely reported.

In conclusion, we reported the effectiveness of landiolol for arterial tachyarrhythmia in a pediatric patient.

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CONFLICT OF INTEREST

The authors declare no conflict of interest for this article.

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